



THE UNIVERSITY *of* EDINBURGH

This thesis has been submitted in fulfilment of the requirements for a postgraduate degree (e.g. PhD, MPhil, DClinPsychol) at the University of Edinburgh. Please note the following terms and conditions of use:

This work is protected by copyright and other intellectual property rights, which are retained by the thesis author, unless otherwise stated.

A copy can be downloaded for personal non-commercial research or study, without prior permission or charge.

This thesis cannot be reproduced or quoted extensively from without first obtaining permission in writing from the author.

The content must not be changed in any way or sold commercially in any format or medium without the formal permission of the author.

When referring to this work, full bibliographic details including the author, title, awarding institution and date of the thesis must be given.



THE UNIVERSITY
of EDINBURGH

**COGNITIVE FACTORS AND SUBJECTIVE WELLBEING
IN PARENTS WHO HAVE CHILDREN WITH
PROFOUND AND MULTIPLE INTELLECTUAL DISABILITY**

Fleur-Michelle Marguerite Coiffait

*Submitted in part fulfilment of the degree of
Doctorate in Clinical Psychology
at the University of Edinburgh*

August 2012

LIST OF CONTENTS

Declaration of own work	5
Epigraph	6
Overview of thesis	7
Abstract	8
Acknowledgements	9
Chapter 1: Systematic Review	10
Abstract	11
Introduction	12
Method	16
Results	27
Discussion	38
Declaration of interests	42
References	42
Chapter 2: Introduction to study	47
Rationale and aims	50
Hypotheses	51
Chapter 3: Journal Article	52
Abstract	53
Introduction	54
Aims and hypotheses	57
Method	57
Results	62
Discussion	63
Summary and conclusions	69
References	69
Chapter 4: Extended Method	74
Ethical considerations	74
Design	74
Participants	75
Procedure	79
Measures	79
Planned data analysis	82

Chapter 5: Extended Results	83
Exploration of data	83
Inferential analyses	86
Summary of results	88
 Chapter 6: Extended Discussion	 89
Clinical implications	96
Strengths and limitations of study	99
Directions for future research	103
Summary and conclusions	104
 References	 106

LIST OF TABLES

Table 1: Study eligibility criteria for the systematic review	18
Tables 2.1 - 2.5: Overview of included studies	22
Table 3: Overview of excluded studies	27
Table 4: Quality assessment ratings for each study	29
Table 1 ^a : Summary of caregiver and child demographic characteristics	59
Table 2 ^a : Key variable summary statistics	62
Table 3 ^a : Multiple regression analysis to predict parental subjective wellbeing	63
Table 5: Demographic characteristics of caregivers	77
Table 6: Health conditions of caregivers	78
Table 7: Demographic characteristics of children with PMID	78
Table 8: Medical conditions and presenting problems of children with PMID	78
Table 9: Key variable measures of central tendency, dispersion, skewness and kurtosis	83
Table 10: Bivariate and point biserial Pearson correlations between key variables	87
Table 11: Multiple regression analysis to predict parental subjective wellbeing	88

^a Tables included in journal article and numbered according to author guidance

LIST OF FIGURES

Figure 1: Flow chart of systematic review study selection process	20
Figure 2: Histogram showing frequency distribution of WEMWBS total scores	84
Figure 3: Histogram showing frequency distribution of PLOC-R total scores	84
Figure 4: Histogram showing frequency distribution of PGS total scores	85
Figure 5: Histogram showing frequency distribution of transformed PGS scores	86

LIST OF APPENDICES

Appendix 1: Author guidelines for journal Research in Developmental Disabilities	112
Appendix 2: Systematic review protocol	119
Appendix 3: List of search terms used in systematic review	122
Appendix 4: Systematic review data extraction form	123
Appendix 5: Quality rating tool for systematic review	125
Appendix 6: Participant information	130
Appendix 7: List of organisations that supported the study	133
Appendix 8: Parental Locus of Control Scale - revised version	134
Appendix 9: Positive Gain Scale	137
Appendix 10: Warwick-Edinburgh Mental Wellbeing Scale	138



THE UNIVERSITY
of EDINBURGH

**D. CLIN. PSYCHOL.
UNIVERSITY OF EDINBURGH / NHS (SCOTLAND) TRAINING PROGRAMME**

Declaration of own Work

NAME: Fleur-Michelle Coiffait

ASSESSED WORK: Doctoral thesis

TITLE OF WORK: Cognitive factors and subjective wellbeing in parents who
have children with profound and multiple intellectual disability

I confirm that all this work is my own except where indicated, and that I have:

- Read and understood the Plagiarism Rules and Regulations
- Composed and undertaken the work myself
- Clearly referenced/listed all sources as appropriate
- Referenced and put in inverted commas any quoted text of more than three words (from books, web, etc)
- Given the sources of all pictures, data etc. that are not my own
- Not made undue use of essay(s) of any other student(s) either past or present (or where used, this has been referenced appropriately)
- Not sought or used the help of any external professional agencies for the work (or where used, this has been referenced appropriately)
- Not submitted the work for any other degree or professional qualification except as specified
- Acknowledged in appropriate places any help that I have received from others (e.g. fellow students, technicians, statisticians, external sources)
- Complied with other plagiarism criteria specified in the Programme Handbook
- I understand that any false claim for this work will be penalised in accordance with the University regulations

Signature: _____ Date: 01/08/2012

EPIGRAPH

Welcome to Holland

(Kingsley, 1987)

When you're going to have a baby, it's like planning a fabulous vacation trip - to Italy.

You buy a bunch of guide books and make your wonderful plans.

The Coliseum. The Michelangelo David. The gondolas in Venice.

You may learn some handy phrases in Italian. It's all very exciting.

After months of eager anticipation, the day finally arrives.

You pack your bags and off you go. Several hours later, the plane lands.

The stewardess comes in and says, "Welcome to Holland."

"Holland?!?" you say. "What do you mean Holland??"

I signed up for Italy! I'm supposed to be in Italy. All my life I've dreamed of going to Italy."

But there's been a change in the flight plan.

They've landed in Holland and there you must stay.

The important thing is that they haven't taken you to a horrible, disgusting, filthy place,
full of pestilence, famine and disease. It's just a different place.

So you must go out and buy new guide books. And you must learn a whole new language.

And you will meet a whole new group of people you would never have met.

It's just a different place. It's slower-paced than Italy, less flashy than Italy.

But after you've been there for a while and you catch your breath, you look around....

and you begin to notice that Holland has windmills....and Holland has tulips.

Holland even has Rembrandts.

But everyone you know is busy coming and going from Italy...

and they're all bragging about what a wonderful time they had there.

And for the rest of your life, you will say

"Yes, that's where I was supposed to go. That's what I had planned."

And the pain of that will never, ever, ever, ever go away...

because the loss of that dream is a very very significant loss.

But... if you spend your life mourning the fact that you didn't get to Italy,
you may never be free to enjoy the very special, the very lovely things ... about Holland.

OVERVIEW OF THESIS

This thesis follows a portfolio format and constitutes part-fulfilment of the academic component of the degree of DClínPsychol at the University of Edinburgh. Other academic components completed over the duration of the author's DClínPsychol studies include nine essays, four case studies and two small scale research projects.

An abstract provides a summary of the entire portfolio thesis, including aims, findings and implications. Chapter One contains a systematic review of published research investigating locus of control and its relevance to the psychological outcomes of parents of children who have a disability. This review was prepared for submission to the journal *Research in Developmental Disabilities*.

Chapter Two links the systematic review to the rationale, aims and hypotheses for the research study. The research study has been written up in the format of a journal article, in preparation for submission to *Research in Developmental Disabilities*. This is presented in Chapter Three. Chapter Four describes the method of the study in further detail and Chapter Five provides extended information regarding the results, including data exploration and inferential statistical analyses.

Chapter Six contains a detailed discussion of the study's findings within the context of the existing literature, explores the implications for clinical practice and discusses limitations of the study and directions for future research. The final sections of the thesis portfolio comprise references and appendices.

The thesis portfolio will adopt the British Psychological Society's editorial style (BPS, 2004) throughout. The systematic review and journal article chapters will be exceptions, where the referencing style of the American Psychological Association (2009) will be used in line with the requirements for the journal *Research in Developmental Disabilities*.

PORTFOLIO THESIS ABSTRACT

Aims: The aims of this thesis were twofold. First, to review the literature on parental locus of control and its role in psychological outcomes for parents who have a child with an intellectual disability (ID). Second, a research study aimed to explore levels of parental subjective wellbeing in a specific group of these parents: those who have a child with profound and multiple intellectual disabilities (PMID). More specifically, whether two different types of parental cognition, parental locus of control and recognition of positive gains of having a child with PMID, were predictive of parental subjective wellbeing.

Method: A systematic review of the literature was conducted to address the first aim. For the research study, a single sample of parents and family caregivers (n=101) completed three quantitative self-report questionnaires as part of a within-participant, cross-sectional survey design. These included the Positive Gain Scale, a modified version of the Parental Locus of Control Scale, and the Warwick-Edinburgh Mental Wellbeing Scale .

Results: The systematic review highlighted the influence of parental locus of control and other parental cognitions on parent and family psychological outcomes. The research study revealed that parental subjective wellbeing in this group of parents (N= 101) was lower than in the general population. Multiple regression analysis revealed that parental locus of control significantly predicted parental subjective wellbeing ($\beta = -.279$, $t(2,99) = 9.419$, $p = .005$), accounting for around 8% of the variance in WEMWBS scores, $R^2 = .081$, $F(2,99) = 5.474$, $p = .006$.

Conclusions and implications: Although the systematic review and the research study highlighted the importance of parental locus of control for parents of children with ID, the results of the study suggest that other factors are also involved in influencing subjective wellbeing of parents of children with PMID. They also indicate a potential role for psychological intervention for parents and families with a focus on adjusting beliefs and expectations and promoting an internal parental locus of control. However, further research exploring the emotions and experiences of this group of parents is needed.

Keywords profound, multiple, intellectual, developmental, disability, parent, carer, cognition, appraisal, wellbeing, adjustment, adaptation, coping

ACKNOWLEDGEMENTS

First, I would like to convey my deepest thanks to all of the parents and caregivers who took part in the study and shared their thoughts and experiences of having a child with profound and multiple intellectual disabilities. It was the courage and resilience of this group of parents that I had the privilege to witness in my clinical work that motivated me to undertake the systematic review and research that forms this thesis. I hope to use these insights to contribute to future research and the development of interventions and services in this field to facilitate the wellbeing of these families. I would also like to extend my gratitude to the numerous fantastic charities and organisations that disseminated details of my study and helped me recruit parents and caregivers, without whom my research would not have been possible. A full list of these can be found in Appendix 7.

I would like to thank my supervisors, Dr Helen Downie and Dr Karen McKenzie, who have both guided and encouraged me throughout the process of completing my thesis. Karen provided invaluable advice and direction and Helen has been extremely supportive, positively influencing both my professional and personal development over the past four years. I would also like to thank Professor Dave Peck for his statistical advice and Becky Wood for kindly giving up her free time to blind-score the papers included in my systematic review.

I am eternally grateful to my wonderful friends and colleagues for their infinite patience, cups of tea and chocolate! Hannah, Amy, and too many others to mention: you made me laugh when I felt like crying and picked me up when I was down. I'd like to thank Dave for taking time out of his own PhD write up to proof read for me and for his salutary advice. Thank you also to Matt, without whom I wouldn't have made it this far. To my family, especially my parents, your unwavering love and belief in me have kept me going. Last, but by no means least, a huge thank you to James for all your love, patience and support over the past year and for your Excel expertise. I couldn't have done this without you all.

I dedicate this thesis to my grandparents, who I have to thank for my drive, determination and desire to discover things.

CHAPTER 1: SYSTEMATIC REVIEW

This chapter contains a systematic review of the research literature exploring the relationship between parental locus of control and psychological outcomes of parents who have a child with a disability. An abstract summarises the findings, followed by background information relating to the area being reviewed, details of the review method and criteria of selection, a summary and discussion of the results, and conclusions and references. The systematic review was prepared for submission to the journal *Research in Developmental Disabilities*, therefore formatting and references follow APA (2009) style. The author guidelines for this journal are provided in Appendix 1.

SYSTEMATIC REVIEW ABSTRACT

A systematic review of the relationship between parental locus of control and psychological outcomes for parents who have a child with a disability

Background: A number of models have been proposed to explain factors associated with parental adjustment and wellbeing, which highlight the importance of social support, parental coping style, and parental appraisals of their situation.

Objectives: This review systematically examined the available literature on a specific type of parental cognition: parental locus of control. The review focused on studies that investigated the relationship between parental locus of control and the psychological outcomes of parents who have a child with a disability, with the aim of informing future research and clinical interventions.

Method: Online database searches and hand searches of three journals in the field led to the identification of ten papers eligible for review. These were assessed against predefined criteria and the findings synthesised.

Results: The ten quantitative studies appraised were assessed as being of fair to good quality. Statistically significant relationships were revealed between parental locus of control and parental stress, anxiety, depression, self-esteem, coping styles, satisfaction and physical incapacitation of the child for parents of children with a disability. Parental locus of control did not have a statistically significant influence on psychological outcomes in two of the studies reviewed.

Conclusions: Parental locus of control had a significant influence on parent psychological outcomes in eight of the ten of studies. The wider importance of parental cognitions beyond parental locus of control was also highlighted, in addition to issues of conceptual overlaps and the dearth of psychometrically robust measures of parental locus of control.

Highlights

- Research studies of parents are often biased towards samples of mothers.
- Current measures of parental locus of control have limitations.
- Parental locus of control overlaps conceptually with other parent cognitions.
- Parental locus of control influences outcomes for parents of disabled children.
- Further research is needed in this area.

Keywords disability, parent, locus of control, wellbeing, adjustment, coping

1. Introduction

1.1 Background

A significant body of research has highlighted that parenting a child with additional needs is associated with higher levels of parental stress, anxiety and depression, and lower levels of parent wellbeing (Davis, 1993; Edwards & Titman, 2010). This finding has been replicated among various groups of parents whose children have additional needs, including those arising from chronic physical illness (Kuster & Badr, 2006), life-limiting illness (Fotiadou, Barlow, Powell & Langton, 2008), genetic conditions (Foster, Kozachek, Stern & Elsea, 2010), physical disability (Ketelaar, Volman, Gorter & Vermeer, 2008), developmental disability (Singer, 2006), and behavioural difficulties (Neece, Green & Baker, 2012). These factors all place increased care burden on parents, either from a practical point of view (e.g. medical care regimes) or an emotional point of view (e.g. feelings of worry, guilt or failure), or indeed a combination of both (McCann, Bull & Winzenberg, 2012; Wallander & Varni, 1998).

Not only does having a child with additional needs impact directly on caregivers, it can also have an indirect effect on relationships with partners, family members and wider social networks, career prospects, and even physical health (Davis, Shelly, Waters, Boyd, Cook & Daverm, 2009; Wallander & Varni, 1998). These sequelae in turn can have a compounding detrimental impact on caregivers and the rest of the family and can make caring for a child with additional needs an extremely challenging role. The daily and cumulative life stressors associated with having a child with additional needs, rather than disease or disability characteristics themselves, have been strongly associated with parental psychosocial functioning (Wallander & Varni, 1998). However, a sizable proportion of caregivers and families are able to adjust well to the demands and challenges of having a child with additional needs (Goodley & Tregaskis, 2006; Wallander & Varni, 1998). There is also considerable individual variance in adaptation to this role, thus it is important to explore the protective processes that serve to strengthen caregiver and family wellbeing, coping and adjustment (Bristol, 1987; Eiser, 1990).

This article will first outline theoretical models that have been used to understand the relationship between stress and psychological outcomes for parents who have a child with additional needs. It will then focus on an important feature of several of these models: parental cognitions. The literature on a particular type of parental cognition that has been highlighted as influential to adjustment and psychological outcomes - locus of control - will then be examined systematically in relation to the stressor event of having a child with a disability.

1.2 Theoretical models

A number of theoretical models have been developed to explain the relationships between stressors and psychological outcomes such as adjustment, coping and wellbeing (Wallander & Varni, 1998). Several of these have been applied specifically to having a child with additional needs and explore how stressor and environment characteristics and resources mediate the relationship between stressor and outcome, and the processes by which this happens.

1.2.1 Stress-coping model (Lazarus & Folkman, 1984)

This transactional theory of stress and coping is one framework that has been used to explain the process of coping and adjustment to stressors in general and has been applied to many contexts. This model proposed that a stressor results in an individual forming cognitive appraisals of a situation, which can be either primary or secondary. Primary appraisals relate to the personal meaning of an event and whether the individual sees it as positive, negative or neutral. Secondary appraisals concern what can be done about the stressor and an individual's thoughts regarding their capacity to reduce any potential loss, threat or damage. These are both thought to be influenced by an individual's overall appraisal style and the model proposed that these determine the coping strategies employed by an individual. Coping strategies are defined as any effort to manage external or internal demands appraised as negative or challenging. These may be behavioural (e.g. avoidance of a situation) or cognitive (e.g. excessive worrying). Multiple potential outcomes follow the appraisal of a stressor and the consequent coping response, but the model broadly defines these in terms of stress or adaptation. It also highlights a number of other factors that moderate the relationship between stressor and outcome via their effects on cognitive appraisals and coping strategies. These include personality characteristics, self-efficacy and social support, among others.

1.2.2 Stress-coping model for coping with chronic disease (Lazarus, 1991)

The transactional stress-coping model has been adapted specifically for explaining the process of coping with chronic illness as a stressor. The adapted model suggests that the condition characteristics, treatment/regime characteristics and condition-related events are important aspects in terms of stressors. As with the original model, these are posited to result in cognitive appraisals of demands and goals, in addition to an emotional response. Internal and external resources are also identified as influencing coping behaviour, which in turn has psychological, social and physical consequences in this model.

1.2.3 Double ABCX Model (McCubbin & Patterson, 1983)

Using the foundations of the Lazarus and Folkman (1984) stress-coping model and the ABCX Model of family stress (Hill; 1949; 1958), the Double ABCX model was developed as a framework for understanding family stress that also takes into account how families recover and adapt after crisis. In the ABCX model (Hill, 1949; 1958), the stressor is represented as 'A' and it interacts with the family's resources, represented as 'B'. These then interact with the family's appraisal or interpretation of the event, represented as 'C'. A, B and C all then influence 'X' - the outcome in relation to the family. The Double ABCX model was developed in response to longitudinal research by McCubbin and Patterson (1983), in an attempt to predict and explain why some families recover and are better able to adapt following crises. In addition to A,B,C, and X, the Double ABCX model incorporates additional life stressors and strains; psychological, intrafamilial and social resources; changes in the family's definition; family coping strategies; and a range of possible psychological outcomes. The main development from the ABCX model is that it includes both 'precrisis' variables (stressor; existing resources; perception of the stressor) and 'postcrisis' variables (pileup of stressors on top of initial stressor; use of existing and new resources; perception of the stress pileup and resources; coping; adaptation to the postcrisis variables).

1.2.5 Parent Disability-Stress-Coping Model (Wallander, Varni, Babani, DeHaan, et al., 1989)

In this model, associated with Wallander and Varni's (1992) disability-stress-coping model of psychological adjustment in children with a chronic illness, stress is a central component that can be exacerbated or ameliorated by parental coping strategies and social support. It focuses on three types of protective mechanisms: stress processing, intrapersonal and social-ecological mechanisms (Wallander & Varni, 1998). Stress processing involves the appraisal of an experience and implementation of coping strategies to manage this. Much like Lazarus and Folkman's (1984) theoretical framework, the importance of event-specific appraisals is emphasised, as this is thought to influence the type of coping strategy implemented, depending on whether appraisals feature perceptions of threat, loss or challenge. Intrapersonal factors refer to general cognitive and affective patterns of behaviour along various dimensions. These include control orientation, dispositional optimism, self-perception and problem-solving abilities (Wallander & Varni, 1998). The final protective mechanism outlined in the model focuses on the social environment, including the family and wider social networks. This aspect of the model draws on extensive evidence highlighting the strong relationships between parental adjustment to having a child with additional needs and the availability of

practical resources, family support, marital satisfaction, and utilisation of social support networks (Wallander & Varni, 1998).

1.2.6 Summary of key theoretical models of family stress, coping and adjustment

Each of the four key theoretical models outlined has highlighted the multi-dimensional and transactional nature of the process of responding to stress. Key variables identified in the stress process that influence outcome include cognitive, emotional and behavioural responses to the stressor. These can be at the level of an individual, family, or wider system and are partly determined by the resources available. Cognitive appraisals are particularly important, as they determine whether the situation is perceived as positive or negative and within or outside one's control. This then influences the individual or family's emotional and behavioural responses and determines the coping strategies they may or may not employ. Therefore, cognitive appraisals will be the subject of this systematic review. Given the range and diversity of cognitive factors that have been linked to the stress-coping process (e.g. threat appraisal, attribution of causality, perceived locus of control, self-efficacy, hope/optimism), parental locus of control has been selected as the specific focus of the review.

1.5 Locus of control as a concept

The perception of how much control one has over a given situation and how this affects subsequent responses to that and other situations has been an area of interest and relevance to many fields of psychology over several decades (Lefcourt, 1982). The most extensive work in this area was derived from Rotter's (1954) social learning theory, which proposed that an individual's actions could be predicted on the basis of their values, expectations and situational context. Rotter (1954) posited that the potential occurrence of behaviours that satisfy some need is a function of the expectancy that those behaviours will lead to these reinforcements, and the strength of value these reinforcements have.

Following on from Rotter's (1954) ideas, perceived control is defined as the generalised expectancy of internal versus external control of reinforcement, and involves a causal analysis of success and failure (Lefcourt, 1982). A person's interpretation of the cause of their experiences is key, and interest in perceived control originated from observations of psychotherapy patients. Rotter (1966) noted that some individuals adapted their behaviour following new experiences and gained from this, whilst others attributed change to others, rather than their own behaviour or characteristics. Therefore, people's expectancies regarding the controllability of outcomes influenced both the way they behaved and the way they made sense of changes in their experience (James & Rotter, 1958).

1.6 Parental locus of control

A growing research literature has begun to investigate the effect of parents' locus of control, specifically in relation to the experience of parenting, on both child behaviour and parent and family functioning (Lloyd & Hastings, 2009). Parental locus of control has been associated with parental stress (Friedrich, Wilturner & Cohen, 1985), pessimism (Rimmerman, 1991), depression (Dunn, Burbine, Bowers & Tantleff-Dunn, 2001), anxiety (Lloyd & Hastings, 2009) and family adaptation (Henderson & Vandenberg, 1992). Research exploring the nature of parents' locus of control beliefs has revealed that those who felt unable to control their child's behaviour and that their child's needs dominated their life reported higher stress (Hassall, Rose & McDonald, 2005; Jones & Passey 2005). Furthermore, parental locus of control is related to maternal positive parental perceptions of their child, with mothers who felt that they could not control their child's behaviour less likely to report positive appraisals of their child (Lloyd & Hastings, 2009).

1.7 Rationale for the review

Given the important role of cognitions in models of stress, coping and adjustment and the variation in adaptation to the challenging role of parenting a child with a disability, parental locus of control is a key cognitive variable implicated in these processes. Although Hassall and Rose (2005) recently reviewed the literature on parental cognitions and adaptation to the demands of having a child with an intellectual disability (ID), to date there have been no systematic reviews undertaken of the effect of parental locus of control on parent psychological outcomes. Therefore, this paper will systematically review research investigating parental locus of control in parents of children with a disability.

1.8 Aims of review

This review aims to identify and critically appraise the research literature and synthesise the findings of these studies in order to provide a useful overview of the relationship between parental locus of control and parental psychological outcomes. A further aim of the review is to identify cognitive factors that promote positive psychological outcomes in this group of parents, in order to inform further research and clinical practice.

2. Method

2.1 Protocol

A selection protocol was developed prior to undertaking the literature search to include papers that met eligibility criteria. This comprised an outline of the review question, eligibility criteria, population of interest, outcomes of interest, planned search strategy, planned data extraction and quality assessment methods, and the intended method of

synthesising and disseminating the findings. As suggested in guidance for undertaking reviews in healthcare produced by York University's Centre for Reviews and Dissemination (CRD, 2009), the protocol predefined the method and scope of the current systematic review, with the aim of minimising bias and facilitating transparency. The systematic review protocol is provided in Appendix 2.

2.2 Eligibility criteria

Given the limited literature in this area, it was decided that all published controlled, quasi-experimental and observational studies would be included in the systematic review that were in English, with no date restrictions. As the review focused on a particular population, only those studies that included parents or family caregivers of children who had a disability (physical, intellectual or developmental) were selected for the systematic review. Significant visual or hearing impairment were deemed as disabilities given their impact on child and family functioning and family life. Furthermore, studies were only included in the systematic review if they investigated parents' or family caregivers' locus of control in relation to the experience of parenting their child, as opposed to general locus of control or health locus of control. The final criterion for inclusion was that the study measured at least one of the following parental psychological outcomes: wellbeing, adjustment, adaptation, stress or mental health (e.g. depression, anxiety) and investigated the relationship between parental locus of control and the psychological outcome(s). A summary of the eligibility criteria is provided in Table 1.

2.3 Exclusion criteria

Studies where abstracts did not provide sufficient detail to make a decision about whether they met inclusion criteria were excluded, in addition to studies where the abstract was unavailable. Conference proceedings were excluded as it would be difficult to appraise the studies based on this limited information. Duplicate records were also excluded. Google Scholar was used to attempt to trace any alternative versions of abstracts (e.g. electronic full-text versions of dissertations). Qualitative studies were excluded as the review focused on relationships between specific variables and in order to maintain consistency of appraisal method. Studies where locus of control was investigated in relation to aspects other than parenting (e.g. health locus of control) were also excluded.

Table 1: Study eligibility criteria for the systematic review

Inclusion criteria	Exclusion criteria
Published case study, small study, controlled study or non-controlled study	Duplicate record
Sample comprised parents or family caregivers	Conference proceedings
Child had a physical, developmental or intellectual disability, or significant sensory impairment that would constitute a disability	Qualitative study
Locus of control related to experience of parenting investigated as a main variable	Review paper
At least one of following parent or family psychological outcomes measured: wellbeing, adjustment, stress or mental health	Abstract unavailable
Relationship between parental locus of control and parent/family psychological outcome(s) explored	Full-text unavailable
	Insufficient information in abstract to determine sample characteristics or variables investigated
	Locus of control investigated in relation to aspects other than parenting

2.4 Information sources

Systematic searches were undertaken of the Web of Knowledge, Ovid (incorporating Embase, Medline, PsycArticles and PsycInfo) and EBSCO (incorporating CINAHL plus, ERIC, Medline, PsycArticles and Web of Knowledge) online databases. All publication years provided by these databases were included, up until the date of the search conducted, 9 July 2012. Hand searches of the reference list of any review papers identified were undertaken in addition to hand searches of three journals: the Journal of Mental Retardation, the Journal of Intellectual Disability Research and the Journal of Clinical Child Psychology. This was in order to detect any further papers that may have met criteria for the systematic review. These journals were selected on the basis of being the three most frequently cited sources of citations in the articles that met eligibility criteria for the review.

2.5 Literature search strategy

The search included a multi-database keyword search, individual database keyword search and topic/subject heading searches. Variations of the following terms were used: parent; mother; father; carer; disability; and locus of control (a full list can be found in Appendix 3).

2.6 Study selection

Using the eligibility criteria outlined in Table 1, the abstracts of the studies identified from the searches were initially reviewed in order to determine whether they would be included for full-text review. The full-text of articles deemed to meet the inclusion criteria for the review based on their abstract were then reviewed as part of the second stage of screening. Those that still met the inclusion criteria were selected to be part of the final methodological review and appraisal stage. A flowchart based on the PRISMA statement (Moher *et al.*, 2009) provides an overview of the systematic review study selection process and details each stage (Figure 1).

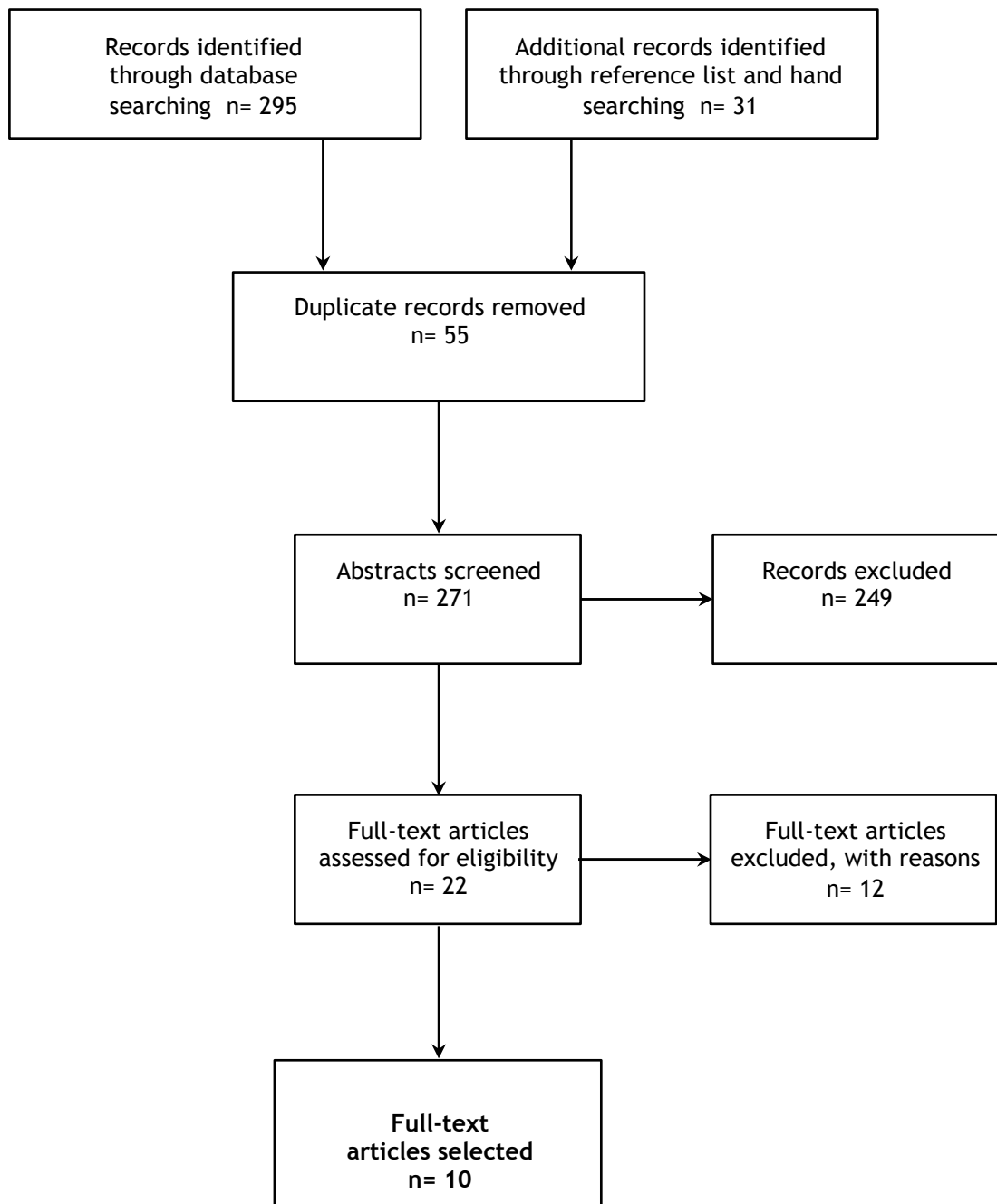
2.7 Data collection

Information was collated for each of the studies included in the final selection for systematic review. This included study characteristics, participant characteristics, and outcome data/results. A standardised form was used for this purpose (see Appendix 4) and a summary of this information is presented for each study in Tables 2.1 - 2.5.

2.8 Assessment of methodological quality

A quality assessment tool was developed for the purpose of assessing and appraising the methodological quality of the studies meeting the inclusion criteria for the systematic review (see Appendix 5). This was based on existing guidelines, including the Scottish Intercollegiate Guidelines Network guidance on systematic literature reviews (SIGN, 2008) and York University's Centre for Reviews and Dissemination guidance for undertaking reviews in healthcare (CRD, 2009).

Figure 1: Flow chart of systematic review study selection process



2.9 Summary measures

Studies were rated using ten quality criteria items across six different dimensions of quality: research questions and objectives; sampling; design and method; statistical analysis, quality of reporting; and generalisability. Numerical ratings were assigned, corresponding to the following quality categories: 1= well covered; 2= adequately addressed; 3= poorly addressed; 0= not addressed/not reported/not applicable. An item was rated as not applicable if it was not relevant to the study design or article. In addition, the overall quality of the study was also rated. This was to avoid a poor score on one index skewing the overall score of an otherwise good quality study. The overall quality ratings assigned were: 3= excellent; 2= good; 1= adequate; 0= poor.

Total numerical scores were calculated for each study, which were then converted to percentages. For items rated as not applicable, percentage calculations were adjusted to reflect the number of applicable items. Finally, percentages were categorised after all articles had been reviewed in order to provide an overall descriptive quality rating for each study (Good $\geq 70\%$, Fair $\geq 50\%$, Weak $< 50\%$). A detailed breakdown of ratings for each study is provided in Table 4.

All studies included in the final selection for review were scored according to the assessment criteria outlined above by two separate researchers, independently of one another. Individual item ratings, sub-totals for each domain and overall scores for each study were assessed for inter-rater reliability. Cohen's Kappa (Cohen, 1960) was good, at .64.

Table 2.1: Overview of included studies

Study	McKinney and Peterson (1987)	Friedrich <i>et al.</i> (1988)
Country of origin	USA	USA
Design	Cross-sectional survey, between-group comparisons by service intervention type and type of diagnosis, plus normative comparisons.	Cross-sectional study, within-group comparisons, with between-group comparisons for demographics.
Method	Self-report postal questionnaires.	Self-report postal questionnaires.
Participants	One group of mothers of children with a diagnosis of Down syndrome, cerebral palsy or other physical disability such as spina bifida.	One group of mothers of children with intellectual disability.
Sampling	N= 67; 32 mothers of children with Down syndrome, 35 mothers of children with cerebral palsy or similar condition such as spina bifida. Recruited from seven early intervention programmes.	N= 140 N= 44 for nonresponder control group used for demographic comparisons.
Variables	Child diagnosis, type of early intervention, social support, perceived control and parenting stress.	Religiosity, parental locus of control, maternal stress, negative wellbeing and depression.
Measures	Parenting Stress Index, Profile of Mood States, Spheres of Control battery, Rotter Locus of Control Scale, Social Network form.	Beck Depression Inventory, Index of Psychological Wellbeing, Religiosity measure, Rotter Locus of Control Scale, Questionnaire on Resources and Stress.
Analyses	Correlational analyses, <i>t</i> tests, ANOVAs, multiple regression analysis.	Two-way ANOVA.
Key results	Spouse support, perceived control and child characteristics accounted for a significant proportion of the variance in stress. Mothers with an internal locus of control and less spouse support had higher stress scores across all measures.	Signification interaction effects between religiosity and child behaviour problems in relation to positive wellbeing; also between religiosity and physical incapacitation regarding positive and negative wellbeing. Significant interaction effects also found between locus of control and physical incapacitation relating to depression. Religiosity appeared to buffer stressors of pessimism, child behaviour problems and physical incapacitation. Locus of control appeared to buffer stressor of physical incapacitation.
Limitations	No discussion of inclusion/exclusion criteria, whether sample characteristic of this population, other confounding variables that may have explained results	No single/unmarried parents included.

Table 2.2: Overview of included studies

Study	Konstantareas and Lampropoulou (1995)	Dunn <i>et al.</i> (2001)
Country of origin	Greece	USA
Design	Cross-sectional survey, between-group comparisons based on median split of single group for main variables.	Cross-sectional survey, within-group comparisons
Method	Semi-structured interviews with researchers	Self-report postal questionnaires.
Participants	One group of hearing mothers of deaf children.	One group of parents of children with a diagnosis of autism.
Sampling	N= 42 Recruited via specialist educational programmes for deaf children.	N= 58; 39 mothers, 19 fathers. Recruited via divisions of Florida Autism Society and Autism Societies of America.
Variables	Stress, child and family characteristics, maternal locus of control, self-esteem.	Parenting stress, in addition to child/parent/family characteristics.
Measures	Questionnaire on Resources and Stress, Rotter Locus of Control Scale, Coopersmith Self-Esteem Inventory.	Inventory of Socially Supportive Behaviours, Ways of Coping questionnaire, Parenting Stress Index, Rotter Locus of Control Scale, Life Experiences Survey.
Analyses	MANOVA, univariate ANOVAs.	Correlational analyses, stepwise regressions, moderator analyses.
Key results	Age of onset of deafness significantly related to maternal stress. Majority of mothers had external locus of control. Self-esteem directly related to stress and best predictor of stress.	Escape-avoidance ways of coping significantly associated with increased depression and social isolation. Confrontive ways of coping significantly associated with decreased depression. Increased use of positive reappraisal corresponded to decreased social isolation. Depression significantly associated with locus of control, distancing and escape ways of coping.
Limitations	Only included mothers.	Minimal information about recruitment, participants' characteristics and variables under investigation. No alternative explanations offered, e.g. confounding variables.

Table 2.3: Overview of included studies

Study	Hassall <i>et al.</i> (2005)	Hamlyn-Wright <i>et al.</i> (2007)
Country of origin	England, UK	England, UK
Design	Cross-sectional survey, within-group comparisons.	Cross-sectional survey, between-group comparisons.
Method	Self-report questionnaires and interviews.	Self-report postal questionnaires.
Participants	One group of mothers of children with intellectual disability.	Three groups of parents: those with children with autism, Down syndrome, or no developmental disability.
Sampling	N= 46 Recruited via special schools.	N= 619; 577 mothers, 42 fathers. Recruited via randomised postal survey to members of the National Autistic Society and National Down Syndrome Association. Group without a developmental disability were recruited via schools (private and local authority).
Variables	Parental cognitions, parenting stress, parental self-esteem and parental locus of control.	Stressors, social support, parental locus of control, coping style, negative outcomes.
Measures	Vineland Adaptive Behaviour Scale, Parenting Stress Index, Family Support Scale, Parenting Sense of Competence Scale, Parental Locus of Control Scale - Short Form, revised.	Goldberg Locus of Control Scale, Hospital Anxiety and Depression Scale, and questions on parental stress drawn from literature.
Analyses	Correlational analyses, stepwise regression analysis.	ANOVAs, post-hoc analyses, mediation analyses with multiple regressions.
Key results	Significant association between child behavioural difficulties and parenting stress, mothers with an external locus of control more likely to experience more stress. Higher self-esteem correlated with more internal locus of control. Regression analysis showed parental cognitive variables (locus of control and parenting satisfaction) were both significant predictors of parenting stress, child behavioural difficulties also significantly contributed to variance in parenting stress.	Significant between-group differences in stress, anxiety, and depression. Parents of children with autism had significantly lower levels of internal locus of control than the two other groups. Locus of control mediated relationship between parental stress and depression/anxiety for parents of children without a developmental disability, but not in the other two groups.
Limitations	Only included mothers, sample not representative of population, however, this is acknowledged in paper.	Impact of these different disabilities on parents not fully explained, stress measure not validated and info on psychometrics not fully provided.

Table 2.4: Overview of included studies

Study	Lloyd and Hastings (2009)	Glenn <i>et al.</i> (2009)
Country of origin	Wales, UK	England, UK
Design	Cross-sectional and longitudinal survey, within-group and longitudinal comparisons.	Cross-sectional study part of larger RCT intervention study for families of children with cerebral palsy. Within-group and normative comparisons
Method	Self-report postal questionnaires.	Self-report questionnaires and researcher assessment of child.
Participants	One group of mothers of children with intellectual disability at time 1; 57 at time 2.	One group of mothers of preschool children (<4) with cerebral palsy (perinatal in origin, predominantly spastic in type).
Sampling	N= 91 Recruited via special schools.	N= 70 Recruited from larger study where families referred via child development centres.
Variables	Parental locus of control and maternal distress (stress, anxiety and depression), child characteristics (adaptive functioning, behavioural problems).	Parenting stress, in addition to child/parent/family characteristics.
Measures	Vineland Adaptive Behaviour Scale, Strengths and Difficulties Questionnaire, Parental Locus of Control Scale, Positive Contributions Scale, Hospital Anxiety and Depression Scale, Questionnaire on Resources and Stress.	Gross Motor Function measure, Griffiths Mental Development Scales, Parenting Stress Index, Family Needs Scale, Family Support Scale, Family Adaptability and Cohesion Scales III, Home Observation for Measuring the Environment, Carver's Coping Scale, Brief Locus of Control Scale.
Analyses	Correlations, linear regression analyses, exploration of longitudinal relationships, then exploratory moderated multiple regression analyses.	Correlation and regression analyses.
Key results	At time 1, maternal positive perceptions of child associated with belief in fate/chance and associated with parental locus of control. Maternal anxiety associated with parental locus of control. Higher external parental locus of control linked with more depression and stress. Bi-directional relationship between maternal stress and parental locus of control. Change in parental locus of control total score across time predicted stress at time 2.	High parenting stress significantly correlated with higher family needs, maladaptive coping, life stressors. Higher parenting stress also significantly correlated with lower family cohesion, family adaptability, Home Observation for Measuring the Environment score, cognitive quotient, in addition to an external locus of control. Family needs, family adaptability and cognitive impairment all significantly predicted overall family stress.
Limitations	Only included mothers, self-selected, high proportion who were well educated and middle class - may affect generalisability.	Little info about sampling provided, insufficient detail provided for replication, limited information about variables investigated.

Table 2.5: Overview of included studies

Study	Siman-Tov and Kaniel (2011)	Lanfranchi and Vianello (2012)
Country of origin	Israel	Italy
Design	Cross-sectional survey, within-group exploration.	Cross-sectional survey, between-group comparisons.
Method	Self-report postal questionnaires.	Self-report postal questionnaires.
Participants	One group of parents of children with diagnosed pervasive developmental disorders (Rett's disorder, childhood disintegrative disorder, Asperger's disorder, or pervasive developmental disorder not otherwise specified).	Four groups of parents of children with diagnosed genetic syndromes (Down syndrome, Williams syndrome, Fragile X syndrome and Prader-Willi syndrome).
Sampling	N=176; 88 married mothers, 88 married fathers. Recruited from parents' associations or community treatment centres.	N= 280; 140 married fathers, 140 married mothers. Recruited from national/local parent organisations, hospitals and rehabilitation centres.
Variables	Resources (sense of coherence, parental locus of control and social support) and stress applied to adjustment (mental health and quality of marriage) to parenting a child with autism.	Parental stress, parental locus of control, family cohesion and family adaptability.
Measures	Questionnaire on Resources and Stress, Sense of Coherence scale, Rotter Locus of Control Scale, Social Support Scale, Mental Health Scale, Quality of Marriage Scale, Autism Behaviour Checklist.	Questionnaire on Resources and Stress, Parental Locus of Control Scale, Family Adaptability and Cohesion Scales III.
Analyses	Structural equation modelling, path analysis.	ANCOVA, post-hoc analyses, multiple regression analyses.
Key results	Sense of coherence, internal locus of control, social support and quality of marriage increased ability to cope with the stress of parenting an autistic child.	Parents of children with Down syndrome experienced significantly fewer parent and family problems and pessimism than other three groups. No significant differences in parental gender found.
Limitations	No discussion of inclusion/exclusion criteria, whether sample characteristic of this population, other confounding variables that may have explained results.	No single/unmarried parents included.

3. Results

3.1 Study selection

A total of 326 records were identified through the literature search (see Figure 1). Figures relating to reasons for exclusion are provided in Table 3. An overview of studies selected for inclusion in the systematic review is provided in Tables 2.1-2.5, followed by a more in-depth summary of the findings relating to parental locus of control and parent psychological outcomes.

3.1.2 Excluded studies

As Table 3 shows, a total of 316 studies were excluded for a range of reasons. These can be broadly categorised as due to duplication, inaccessibility, or populations or methods outside the scope of the review question.

Table 3: Overview of excluded studies

Number of studies excluded	Reasons for exclusion
174	Different subject/research focus
70	Duplicate record
16	Sample included parents of children who did not have a disability or significant sensory impairment/medical condition that would constitute a disability
9	Insufficient information in abstract to determine sample characteristics or variables investigated
9	Not in English
9	Relationship between parental locus of control and parent psychological variables not investigated
7	Locus of control investigated in relation to aspects other than parenting (e.g. general or health locus of control)
7	Parent psychological variables were not investigated
5	Focused on other family members (e.g. siblings, partners)
4	Review paper
2	Qualitative study
2	Abstract unavailable
2	Sample included parents of adults with a disability
0	Full-text unavailable
0	Conference proceedings
316	Total number of studies excluded

3.1.3 Included studies

As summarised in Tables 2.1-2.5, ten quantitative studies undertaken in six different countries between 1987-2012 were selected for appraisal and review of their methodological quality (Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001; Friedrich, Cohen, & Wilturner, 1988; Glenn, Cunningham, Poole, Reeves, & Weindling, 2009; Hamlyn-Wright, Draghi-Lorenz, & Ellis, 2007; Hassall, Rose, & McDonald, 2005; Konstantareas & Lampropoulou, 1995; Lanfranchi & Vianello, 2012; Lloyd & Hastings, 2009; McKinney & Peterson, 1987; Siman-Tov & Kaniel, 2011). These included a total of 1522 parents (1233 mothers, 289 fathers) of children with developmental or genetic disorders; intellectual or physical disabilities; or significant hearing impairment labelled as 'deafness'. In order to answer the systematic review question regarding the relationship between parental locus of control and parental psychological outcomes, all ten studies selected investigated parental locus of control as an independent or predictor variable. A range of parental psychological variables were also investigated in addition to parental locus of control in these studies, usually as dependent variables.

Parental stress was the most frequently studied parent psychological outcome. All ten studies included parental stress as a variable, most commonly measured using the Parenting Stress Index (PSI). Parental anxiety, depression, general distress, general mental health, and wellbeing were also variables investigated in the studies reviewed. In terms of more global parent characteristics, sense of coherence and parental self-esteem were also explored, in addition to ways of coping, religiosity and marital satisfaction. Child characteristics that were investigated included autistic features, emotional and/or behaviour problems, motor function, developmental stage, adaptive function, and child characteristics associated with stress on the PSI. Family cohesion, and adaptability were also investigated, and demographic characteristics were examined across all ten studies. Nine out of the ten studies adopted a cross-sectional design, the remaining study was longitudinal with data collected at two time points.

3.2 Methodological quality of studies

A summary of each paper's methodological ratings on each domain is provided in Table 4, in addition to total scores, percentage scores and corresponding methodological quality category descriptors.

Table 4: Quality assessment ratings for each study

		Hamlyn-Wright <i>et al.</i> (2007)	Konstantareas and Lampropoulu (1995)	Lanfranchi and Vianello (2012)	Siman-Tov and Kaniel (2011)	Hassal <i>et al.</i> (2005)	Lloyd and Hastings (2009)	Green <i>et al.</i> (2009)	Friedrich <i>et al.</i> (1988)	Dunn <i>et al.</i> (2001)	McKinney and Peterson (1987)
Percentage and overall quality category		84.8 Good	78.8 Good	78.8 Good	78.8 Good	72.7 Good	72.7 Good	66.7 Fair	66.7 Fair	57.6 Fair	57.6 Fair
Total	max= 33	28	26	26	26	24	24	22	21	19	19
Overall methodological quality rating	max= 3	3	2	2	2	2	2	2	2	2	2
Generalisability	max= 3	3	2	2	2	2	2	2	1	2	1
Quality of reporting	max= 3	3	2	3	2	2	2	2	3	2	2
Statistical analysis	max= 3	3	2	3	3	2	3	3	2	3	2
Design and method	max= 9	8	6	7	8	7	7	6	6	6	5
Sampling	max= 9	6	9	6	6	6	6	5	5	2	5
Research questions and objectives	max= 3	2	3	3	3	3	2	2	2	2	2

3.2.1 Research question and objectives

All ten studies addressed an appropriate and clearly focused question, drawn from a theoretical model or previous research highlighting the role of locus of control in psychological outcomes for parents and families in the population studied. Theoretical models used as the basis for the research question included the Mash and Johnston (1990) model of determinants of parenting stress; the double ABCX model of family stress (McCubbin & Patterson, 1983); and the transactional model of stress and coping (Lazarus & Folkman, 1984). All ten studies contextualised the development of their research questions in previous research highlighting the increased demands of having a child with additional needs or disability and factors that had been associated with variation in parental psychological outcomes such as adjustment, stress, overall distress, general mental health, anxiety and depression. These factors can be broadly summarised as child factors (behaviour problems; functional impairment; characteristics of condition); parental factors (cognitions, coping style, relationship satisfaction, self-esteem, self-efficacy); and environmental factors (available services; social supports).

3.2.2 Sampling

The representativeness of the samples selected for each study varied, with six of the ten studies only including mothers in their samples (Friedrich *et al.*, 1988; Glenn *et al.*, 2009; Hassall *et al.*, 2005; Konstantareas & Lampropoulou, 1995; Lloyd & Hastings, 2009; McKinney & Peterson, 1987). In those studies that included fathers, two included samples of married parental couples (Lanfranchi & Vianello, 2012; Siman-Tov & Kaniel, 2011) and in the remaining two studies, no information was provided regarding the participants' marital status (Dunn *et al.*, 2001; Hamlyn-Wright *et al.*, 2007;). Fathers have been found to differ from mothers with regard to the way they think about, make sense of and adjust to having a child with additional needs (Hastings, Beck, & Hill, 2005; Hastings, Kovshoff, Brown, Ward, Espinosa, & Remington, 2005; Janssens, 1994; Lanfranchi & Vianello, 2012) and this was the justification for inclusion of mothers only in several studies. However, fathers continue to be a group underrepresented in research on parents (Phares, Fields, Kamboukos, & Lopez, 2005).

The studies reviewed also varied in terms of how much information was provided regarding numbers of potential participants approached, numbers who actually took part and numbers who declined to take part or dropped out. Around half of the studies provided this information, although due to the recruitment methods (e.g. via parent support organisations) it was not always possible to determine this. One study looked at the characteristics of a sample of non-responders in some detail in order to ascertain whether their sample was representative of the population it was drawn from (Friedrich *et al.*, 1988). This highlighted that the responder sample was significantly more educated than the random sample of non-responders, which may have had implications for the results and

conclusions (Friedrich *et al.*, 1988). This is a useful method of not only determining the generalisability of the findings, but also exploring the characteristics of parents who do not take part in research, in order to try and capture the reasons why this may be the case and make future studies more accessible and representative.

Descriptions of the exact nature and severity of children's disabilities or impairments and the impact on everyday family and child functioning varied across the studies. Even in those studies that defined the additional needs of the child related to their condition or disability explicitly, very few studies elaborated on how this impacted on the child and family's everyday functioning. This information serves to contextualise any condition specific-factors that may account for parental and family psychological outcomes and can also explain why parents may perceive that they have more or less control in their caring role.

3.2.3 Design and method

All of the studies except one (Lloyd & Hastings, 2009) adopted a cross-sectional survey design, making the directionality of relationships difficult to infer (Hamlyn-Wright *et al.*, 2007). Of the ten studies, only one included a comparison group of parents of children without additional needs (Hamlyn-Wright *et al.*, 2007), although three studies did include between-group comparisons using sub-groups of the sample. Lanfranchi and Vianello (2012) compared the results of parents who had children with four different types of genetic condition with differing characteristics. McKinney and Peterson (1987) based their within-group comparisons on the type of service intervention participants were receiving, and Konstantareas and Lampropoulou (1995) used a median-split of the scores on their main variable of interest to make within-group comparisons.

Self-report questionnaires were distributed via post in all studies but one, where researchers went through questionnaires with participants as part of a semi-structured interview (Konstantareas & Lampropoulou, 1995). Administering measures in person is arguably more time-consuming, but may enable more representative data to be gathered from those who may not understand the questionnaires or who need support to complete them. These individuals might otherwise fail to complete or return questionnaires distributed via post, potentially leading to a bias towards inclusion of responses of individuals who may, for example, have more time or may be more educated or motivated to return their responses, such as in the Friedrich *et al.* (1988) study.

Reliance on data gained solely from parental report also increases the possibility of source variance and the identification of associations based on this, rather than true effects (Hassall *et al.*, 2005). Bugental, Johnston, New, and Silvester (1998) expressed concerns relating to parental impression management when completing self-report questionnaires

about their parenting. This is difficult to avoid when the subject of the research concerns parental experiences and perceptions.

3.2.4 Statistical analysis

A range of statistical analyses were employed in the ten studies reviewed (see Tables 2.1-2.5 for details). Analyses in all of the studies were appropriately reported and justified, with some providing more extensive detail than others (e.g. a description of how parametric assumptions were explored).

3.2.5 Quality of reporting

Similarly, the quality of reporting across all of the ten studies reviewed was judged to be adequate or good. Again, some studies provided more detail than others, but this appeared to be related to the article length requirements of the different journals. Older studies tended to include less detail, which may have been due to restricted print space or accepted protocol at the time, although this is unclear.

3.2.6 Generalisability

The generalisability of the papers reviewed ranged from poor to good, with the findings of the majority of studies reviewed rated as adequate. It was difficult to assess generalisability for some studies as inadequate information was provided about the sample. The main factors that undermined generalisability were the dearth of fathers included in the studies, the very specific populations studied and the tendency for most samples to be recruited via parent support organisations. There may have been a self-selection bias, whereby parents experiencing more difficulties were more likely to take part (Hamlyn-Wright *et al.*, 2007). It may also be that those parents who do not engage with support organisations are coping effectively, or it may be that they are experiencing a higher rate of negative outcomes (Dunn *et al.*, 2001). The findings of these studies should therefore be generalised with caution as they may not represent the experiences of all parents in that population.

3.3 Synthesis of results

3.3.1 Parental psychological outcomes

In terms of the parent psychological outcomes investigated in relation to parental locus of control in the studies reviewed, stress was the dominant parental psychological outcome explored, appearing in all ten studies. Parental mental health (Siman-Tov & Kaniel, 2011), coping (Dunn *et al.*, 2001; Glenn *et al.*, 2009), negative and positive wellbeing (Friedrich *et al.*, 1988) were also measured, in addition to anxiety (Hamlyn-Wright *et al.*, 2007; Lloyd & Hastings, 2009), depression (Friedrich *et al.*, 1988; Hamlyn-Wright *et al.*, 2007; Lloyd & Hastings, 2009), and parental self-esteem (Hassall *et al.*, 2005; Konstantareas &

Lampropoulou, 1995). Parental psychological outcomes directly related to parenting that were investigated included parenting sense of competence, satisfaction and self-efficacy (Hassall *et al.*, 2005). In addition to these parental psychological outcomes, psychological outcomes that were more relational that were explored included quality of marriage (Siman-Tov & Kaniel, 2011), family cohesion and family adaptability (Glenn *et al.*, 2009; Lanfranchi & Vianello, 2012). Parental psychological outcomes that were significantly related to parental locus of control will be discussed in the next section in more detail.

3.3.2 Role of parental locus of control

An internal parental locus of control was associated with lower levels of reported depression and isolation in parents of children with autism (Dunn *et al.*, 2001). However, parental locus of control did not appear to buffer against the negative effects of stress or enhance the positive effects of social support in this particular study. Furthermore, little information was provided regarding those who declined to take part in this study, therefore these results may represent those parents who were particularly motivated to take part because they were coping well, or the reverse. Hassall *et al.* (2005) demonstrated strong evidence of the relationship between cognitive states and parenting stress in mothers of children with ID, who face similar issues to those of parents of children with autism. Strong positive correlations between parental locus of control and parenting stress highlighted that mothers of children with ID who had a more external parental locus of control experienced higher levels of stress (Hassall *et al.*, 2005). However, the generalisability of these findings is also limited due to the fact that samples were drawn from a predominantly middle class, rural area and no information was provided about the characteristics of those who declined to take part.

Lloyd and Hastings (2009) also reported that parental locus of control in mothers of children with ID was significantly associated with stress, as well as with depression and anxiety, in line with previous research (Friedrich *et al.*, 1985). Mothers who had a more external parental locus of control reported more symptoms of distress (Lloyd & Hastings, 2009). Furthermore, the development of a more external parental locus of control across time was associated with increased parental stress (Lloyd & Hastings, 2009). One theoretical explanation put forward for these findings was that mothers who feel that their child's behaviour is beyond their control may develop learned helplessness (Seligman, 1975). Friedrich *et al.* (1988) also proposed that a perception of having some control in a situation mitigates against hopelessness, which they highlight is a common precursor of depression.

Glenn *et al.* (2009) used cluster analysis to group families with similar characteristics and found that those with a more internal locus of control tended to have average to high levels of family cohesion, low to average levels of maladaptive coping, average levels of stressful life events and family support, and children with higher cognitive abilities. Those

with a more external locus of control often had low levels of family support, very low levels of family cohesion, but average levels of child impairment. Parents with a high external locus of control were characterised by high levels of stressful events and maladaptive coping, average levels of family support, low levels of family cohesion and cognitive functioning of their child, and severe levels of child impairment. However, these findings tell us little beyond the types of families emerging in this particular study. Cluster analysis has been questioned in terms of the meaningfulness of the models yielded from the data and is often criticised in terms of how readily one can generalise from these type of results to larger populations (Henry, Tolan, & Gorman-Smith, 2005).

Siman-Tov and Kaniel's (2011) study of parents with developmental disorders also revealed further evidentiary support for a strong link between parental locus of control and parental stress. A more external parental locus of control appeared to be associated with higher levels of stress, but the directional relationship between parental locus of control and other factors investigated is unclear (Friedrich *et al.*, 1988; Hassall *et al.*, 2005). Beliefs related to locus of control may also interact with other coping resources and previous coping experiences to influence parent psychological outcomes (Beresford, 1994; Hassall *et al.*, 2005) and further longitudinal research is needed in this area (Siman-Tov & Kaniel, 2011).

Although eight of the ten studies have highlighted a clear relationship between parental locus of control and parent psychological outcomes, Hamlyn-Wright *et al.* (2007) found that locus of control only mediated the relationship between parental stress and both depression and anxiety for parents of children without a developmental disorder. This was not the case for parents of children with a developmental disorder in their study (Hamlyn-Wright *et al.*, 2007). There could be a number of reasons for these findings differing from the majority of the studies reviewed. Parents were excluded from the study if their child had a diagnosis of a learning disability, although the exact definition used was not provided and the term can be misleading. It is used in America to represent what is generally referred to as a specific learning difficulty here in the UK. However, in the UK, the term learning disability is often used interchangeably with ID. If parents of children with ID were indeed excluded, this would highlight an important difference between this and the other studies reviewed, as the majority included parents of children who had an ID.

Nevertheless, this study also used a measure of parental locus of control that differed from the measures employed in the other studies reviewed and this may account for the different pattern of results observed. The authors selected a general locus of control measure that was part of an inventory of personality tests (Goldberg, 1999; International Personality Item Pool, 2002), stating that they specifically avoided measures such as the Parental Locus of Control Scale as they believed the explicit focus on parenting would

confound its relationship with parenting stress (Hamlyn-Wright *et al.*, 2007). However, such a general 'trait-like' measure as that which appears to have been used may not have been sensitive enough to the more 'state-like' cognitions of parents in relation to the role of parenting a child with additional needs.

In fact, Lloyd and Hastings (2009) noted that the moderate stability co-efficients of parental locus of control scores in their study using the Parent Locus of Control Scale indicated that it may not be a typical trait-like variable and may be affected by the environment in a state-like manner. The relative instability of the Child Control domain of the Parental Locus of Control Scale revealed by Lloyd and Hastings (2009) could be indicative of parents feeling differently about how their child's needs affect their lives at different times. The fact that Hamlyn-Wright *et al.* (2007) utilised a novel measure of stress put together for the purpose of the study and gave little information regarding its psychometric properties also further undermines their findings.

In the Konstantareas and Lampropoulou (1995) study, age of onset of the child's deafness and parental self-esteem were significantly associated with parenting stress. However, parental locus of control was not. The authors noted that 83 per cent of the mothers in their sample were categorised as having an external locus of control and that this may explain the pattern of their results. It is suggested that cultural factors may account for this high proportion of mothers with an external parental locus of control, as the authors outlined that in Greece there is little well organised and predictable support available from services and so mothers may feel like there is a lot outside of their control (Konstantareas & Lampropoulou, 1995). However, without a comparison group, this cannot be elucidated. A further limitation of this study is the exclusion of fathers, although this is acknowledged by the authors who explained that fathers were sought during recruitment but did not wish to participate (Konstantareas & Lampropoulou, 1995).

Despite several limitations in terms of sampling and analysis in the studies reviewed, there was a consensus across eight of the studies that a more external parental locus of control was associated with negative parent psychological outcomes. These included higher levels of depression and isolation (Dunn *et al.*, 2001), parental stress (Hassall *et al.*, 2005; Lloyd & Hastings, 2009) and depression and anxiety (Lloyd & Hastings, 2009). These findings also replicate the previous research of Friedrich *et al.* (1985), thus the relationship between an external parental locus of control and various negative parent psychological outcomes appears to be robust. Furthermore, external locus of control appears to be linked with maladaptive coping, which fits with the above findings.

More detailed analysis of Parental Locus of Control Scale subscale scores revealed that the Child Control and Parent Control subscales were strongly correlated with scores on the

Parenting Stress Index, with the remaining subscales revealing much smaller associations (Hassall *et al.*, 2005). This suggests that these two subscales show the most utility in predicting parenting stress, a finding that replicates previous research (Campis *et al.*, 1986; Hagekull *et al.*, 2001). Lloyd and Hastings (2009) similarly found that the Parental Control subscale correlated significantly with all maternal wellbeing measures used in their study and significantly predicted positive perceptions and anxiety at time one. Therefore, the Parental Control domain of the Parental Locus of Control Scale appeared to be the most useful predictor of parenting stress (Hassall *et al.*, 2005) and maternal wellbeing.

The Parental Responsibility subscale of the Parental Locus of Control Scale was not related to any of the maternal wellbeing variables measured at time one in Lloyd and Hastings' (2009) study. Lanfranchi and Vianello (2012) reported that parents of children with Fragile X syndrome had lower scores on this domain than parents of children with other genetic conditions (Williams syndrome, Down syndrome and Prader-Willi syndrome). It may be that this domain is related to factors that were not fully investigated in these studies, such as the level of services parents were receiving or whether they were actively involved in parent support groups or organisations. Therefore, these findings are inconclusive and it would be useful to include these factors as variables in future research.

Parents of children with Fragile X syndrome and Prader-Willi syndrome scored more highly than other groups of parents with genetic syndromes on the Child Control of Parents' Life domain of the Parental Locus of Control Scale (Lanfranchi & Vianello, 2012). The authors purport that this indicates an external parental locus of control. However, although the Italian short-form of the QRS was used in this study, which included a domain measuring Child's Characteristics, children's levels of behavioural difficulties, adaptive and intellectual functioning were not explicitly measured. This represents a significant limitation as it is likely that the group differences observed in the Lanfranchi and Vianello (2012) study could be alternatively explained in terms of condition sub-group differences on these variables and their interactions with parental locus of control.

Parents who perceived their child as having a higher number of behaviour problems also tended to have a more external parental locus of control and higher levels of stress (Lanfranchi & Vianello, 2012). Inclusion of an objective, other-rated measure of child functioning and behavioural difficulties would have been a useful addition to this study, as it may be that parents who perceive their role as more stressful and who feel they have little control of their situation also perceive their child as being more problematic. It is difficult to unpick the direction of these complex relationships relying on parent self-report measures and a cross-sectional design. This study also reported that the Child Control of Parents' Life domain of the Parental Locus of Control Scale appeared to be particularly important, as it predicted parental stress across all genetic conditions, as did the Child's

Characteristics domain. It seems that parents' perceptions of their child's characteristics and of how much control the child exerts over their parents' lives appear to be key cognitive variables implicated in psychological outcomes of parents whose children have a genetic condition (Lanfranchi & Vianello, 2012).

Rotter (1966) actually highlighted in his original work on locus of control that the repetition of environmental events can weaken or strengthen behavioural expectancies, yet only one of the studies reviewed employed a longitudinal design. Further research of a longitudinal nature is required to elucidate how parental locus of control affects parent psychological outcomes over time and the direction of these relationships. In the only longitudinal study reviewed, Lloyd and Hastings (2009) did not report any significant relationships between parental locus of control and maternal wellbeing across time, but a bi-directional relationship was observed between maternal stress and changes in the total parental locus of control score across time. However, 33 per cent of the sample were lost to follow-up at time two and this may have influenced the results.

It was also posited that the Parental Locus of Control Scale that was used in several of the studies reviewed required further development, due to the several adaptations of the original measure currently in use (Lloyd & Hastings, 2009). It has been reported elsewhere that this measure would benefit from more use with larger samples to ensure reliability (Hagekull *et al.*, 2001). In addition, the discriminant validity of the Parental Locus of Control Scale requires further investigation, as it may be measuring other constructs such as parental distress or other cognitive variables (Lloyd & Hastings, 2009). Thus, the fact that there is currently no psychometrically sound instrument available to measure parental locus of control that is well validated with the populations of parents studied is a limitation of all of the studies reviewed.

It is evident that further research is needed to better understand parental locus of control in parents of children with disabilities, especially fathers, who are underrepresented in the research literature (Olsson & Hwang, 2008). This should ideally be longitudinal in order to elucidate the direction of any emerging relationships and should take into account any factors that may affect parental locus of control, such as the level of support provided to a family from services. It would also be useful to explore locus of control in other carers of children with additional needs, such as carers in the wider family and paid health, social care and education professionals (Lloyd & Hastings, 2009). Once more sensitive, refined measures of parental locus of control are available, it may represent an ideal outcome variable for measuring change following interventions with parents (Lloyd & Hastings, 2009). Lloyd and Hastings (2009) also suggested that it may be fruitful to investigate control in other areas of parents' lives, as this may vary in comparison with feelings of control in the parenting role.

4. Discussion

4.1 Summary of evidence

The ten papers reviewed yielded a number of findings relating to the relationship between parental locus of control and parental psychological outcomes for parents who have a child with additional needs due to a disability, medical condition or severe sensory impairment. The key findings highlighted a strong link between an external parental locus of control and increased negative parental psychological outcomes. These included higher levels of stress, anxiety, depression and general distress, in addition to use of more maladaptive coping strategies. On the other hand, a more internal locus of control was associated with lower levels of parental stress.

Other cognitive factors that influenced parental psychological outcomes included parental sense of coherence, self-efficacy, self-esteem and satisfaction related to their parenting role. These concepts appear to have some overlap with parental locus of control. It became clear that parents' perceptions of factors such as their child's level of impairment or disability and the characteristics of their child's condition were also related to parental stress and coping. Other factors that were identified as being influential to parent psychological outcomes included parental coping style and social support. Finally, the papers reviewed revealed that a number of parents were functioning well and were able to appreciate the positive aspects of having a child with additional needs and this was linked to lower levels of stress and higher levels of wellbeing.

4.2 Theoretical implications

Four key theoretical models were outlined earlier that attempt to explain the relationship between stressors and psychological outcomes, such as adjustment, coping and wellbeing. These were the Stress-coping model (Lazarus & Folkman, 1984), the Stress-coping model for coping with chronic disease (Lazarus, 1991), the Double ABCX model (McCubbin & Patterson, 1983) and the Parent Disability-Stress-Coping Model (Wallander, Varni, Babani, DeHaan, *et al.*, 1989). All of these models propose that individuals may manifest different psychological outcomes in response to similar stressors and that individual variation results from various factors internal and external to the individual coping with the stressor. The studies reviewed were grounded in these theoretical models and investigations focused on identification of particular factors that could account for individual variation. The wider effects of stressors were also acknowledged, with several studies measuring how relational variables, such as family cohesion and marriage quality, were influenced by the stressor of having a child with a disability.

The studies reviewed provided support for several elements of the two stress-coping models developed by Lazarus and colleagues (Lazarus & Folkman, 1984; Lazarus, 1991) and the

Double ABCX model (MCubbin & Patterson, 1983). Stressor characteristics, conceptualised in this case as aspects of the child's disability, were identified as being important to parent psychological outcomes. Studies highlighted that a diagnosis of autism was associated with higher levels of parental stress (Hamlyn-Wright *et al.*, 2007; Lanfranchi & Vianello, 2012), a finding consistent across the wider literature (International Association for the Scientific Study of Intellectual and Developmental Disabilities, 2012). Furthermore, other child characteristics such as their level of physical incapacitation (Friedrich *et al.*, 1988; McKinney & Peterson, 1987), developmental disability (McKinney & Peterson, 1987), behavioural difficulties (Hassall *et al.*, 2005; Konstantareas & Lapropoulou, 1995) and the age of onset of their disability (Konstantareas & Lapropoulou, 1995) all influenced parental psychological outcomes. According to the models mentioned, this may be due to the inherent nature of the stressor characteristics or to the fact that they are appraised by the parent as challenging.

All four theoretical models highlight the key role of appraisals of the stressor and how equipped the individual feels to deal with it. Six of the ten studies identified various types of parental cognitions (e.g. locus of control) as significantly correlated with psychological outcomes, including parental stress, depression and coping (Glenn *et al.*, 2009; Hamlyn-Wright *et al.*, 2007; Hassall *et al.*, 2005; Lloyd & Hastings, 2009; McKinney & Peterson, 1987; Siman-Tov & Kaniel, 2011), providing evidentiary support for this element of the models. However, there was also evidence that parental cognitions are influenced themselves by other factors, such as the quality of marital and family relationships (Glenn *et al.*, 2009; McKinney & Peterson, 1987; Siman-Tov & Kaniel, 2011), religiosity (Friedrich *et al.*, 1988) and even the wider cultural and political context (Konstantareas & Lapropoulou, 1995). Furthermore, based on the literature reviewed, these are likely to be bi-directional relationships. For example, Lloyd and Hastings (2009) identified one such relationship between maternal stress and parental locus of control, and Dunn *et al.* (2001) found that increased parental positive reappraisals corresponded to decreased social isolation. However, the cross-sectional nature of the majority of these studies makes it difficult to determine the direction and nature of causality in these relationships.

Although anxiety and depression were measured across several of the studies, this was often directly linked to parental stress. The range of emotions experienced in response to the stressor of having a child with a disability requires further attention in both the research literature and theoretical models. Qualitative research exploring the nature of parental experiences would inform this and would add to current theoretical conceptualisations of the stress process that emphasise cognitive factors, given the well documented link between cognitions and emotions.

The Double ABCX model emphasises the fact that having a child with a disability is a stressor that unfolds and changes over time, as do families' resources and responses. The only longitudinal study reviewed revealed that if mothers developed a more external parental locus of control across time, this was associated with increased parental stress (Lloyd & Hastings, 2009). Thus there is a clear need for research that monitors the stress coping trajectory of families over time in order to fully understand it.

4.3 Implications for practice

Practitioners working with families of children with additional needs must remain aware of the variety of beliefs that parents may hold (Friedrich *et al.*, 1988). The potential for parental beliefs to be a coping resource has been emphasised, but the need to mobilise other supports and resources has also been highlighted (Friedrich *et al.*, 1988). Dunn *et al.* (2001) suggested that facilitating social support and adaptive ways of coping amongst parents of children with additional needs would be beneficial, in addition to enhancing parents' sense of control over their situation. Furthermore, their findings regarding the unhelpfulness of emotion-focused escape and avoidance coping styles indicates that practitioners should discourage the use of this type of strategy, including hoping for miracles, having fantasies, avoiding contact with others or using food or substances as avoidance.

Encouraging use of a range of coping styles is important, especially positive appraisal and confrontive coping, as these have been found to be more likely to lead to positive parent psychological outcomes (Dunn *et al.*, 2001). Positive reappraisal includes rediscovering the important things in life, finding new faith and meaning, growing as a person and being inspired to be more creative (Dunn *et al.*, 2001). Confrontive coping includes fighting for what is wanted, letting feelings out, expressing appropriate anger towards the problem and taking chances (Dunn *et al.*, 2001). Hassall *et al.* (2005) added that existing behavioural interventions offered by services that aim to reduce children's difficult behaviour also need to include components that address parental coping styles and beliefs regarding their own parenting capacities, in the context of the demands their child presents that may be attributed to causes outside of their control.

Investigation of the relationships between parental locus of control, parenting satisfaction and adaptive coping styles would be useful for informing the development of interventions for parents to ameliorate the stress they experience in relation to parenting a child with additional needs (Hassall *et al.*, 2005; Weiss, 2002). However, Hamlyn-Wright *et al.* (2007) encourage further research exploring parental locus of control that involves interventions designed to directly decrease the stress associated with parenting a child with additional needs. They suggest that interventions aimed at increasing parents' perceived control over their environment are unlikely to influence levels of depression or anxiety in parents who

have a child with additional needs, although this may be useful for parents who have typically developing children (Hamlyn-Wright *et al.*, 2007). Instead, they suggest that psychological interventions for parents of children with additional needs should focus on increasing acceptance of external stressors independent of the child-parent relationship that may be uncontrollable. They also posit that additional help with these stresses and the practical difficulties experienced by parents, rather than attributional retraining, is likely to be beneficial to promote parental adjustment (Hamlyn-Wright *et al.*, 2007).

4.4 Strengths and limitations of review

This is the first systematic exploration of the relationships between parental locus of control and psychological outcomes for parents who have a child with a disability. It offered a particular focus on implications for practice for those working with this group of families. However, the review has a number of limitations that mean the results and conclusions may not be readily generalisable beyond the populations studied. The samples of parents included in the studies reviewed had considerable heterogeneity, although this could also be considered a strength as the findings seemed to converge across different groups of parents with children with various disabilities and conditions.

Due to resource constraints, the review excluded studies that were not published in English, thus introducing a bias for research conducted in English-speaking, Western cultures. Parental locus of control may manifest differently in other cultural settings and it is important to examine a broad range of parenting experiences and practices in order to identify protective factors. Furthermore, the samples included in the papers reviewed were predominantly Caucasian, middle-class, married parents of high socio-economic status, thus the findings may not be generalisable to parents in different demographic circumstances.

Finally, qualitative research was excluded from the review in order to maintain consistency of methodological appraisal. Considering the importance of the nature of parents' thoughts, attitudes, perceptions and appraisals of their family circumstances, it would be informative to explore qualitative investigations of the nature of parents' thoughts, beliefs and experiences when they have a child with a disability.

4.5 Conclusions

Research investigating the role of parental cognitive factors and parental psychological outcomes for parents of children with a disability is steadily gathering pace. A number of good quality cross-sectional questionnaire based studies have been conducted over the past two decades that provide evidentiary support for the utility of existing theoretical concepts and frameworks. The majority of studies reviewed highlighted the relationship between an external parental locus of control and a variety of negative parental psychological outcomes. There is now a need to build on these exploratory findings to elaborate the

mechanisms at play when experiencing the stressor of caring for a child with a disability and the psychological sequelae for the child, parent, family and wider systems. Good quality research in this area will enable the development of an evidence base that will in turn inform the development of effective interventions that can be used in practice to enhance the psychological wellbeing of those caring for a child with a disability and thus promote the wellbeing of the child and wider family.

4.6 Declaration of interests

The primary author conducted the systematic review as part of a portfolio thesis for a Doctorate in Clinical Psychology undertaken at the University of Edinburgh. This training place was jointly funded by NHS Education Scotland and NHS Lothian health board. No other sponsorship, funding or support was received.

References

- Beresford B. A. (1994) Resources and strategies: How parents cope with the care of a disabled child. *Journal of Child Psychology and Psychiatry*, 35,171-209.
- Bristol, M. M. (1987). Mothers of children with autism of communication disorders: Successful adaptation and the Double ABCX Model. *Journal of Autism and Developmental Disorders*, 17(4), 469-486.
- British Psychological Society (2000) *Learning disability: Definitions and contexts*. Leicester: Author.
- Bugental, D. B., Johnston, C., New, M., & Silvester, J. (1998). Measuring parental attributions: Conceptual and methodological issues. *Journal of Family Psychology*, 12, 459-480.
- Campis L. K., Lyman R. D., & Prentice-Dunn, S. (1986). The Parental Locus of Control Scale: Development and validation. *Journal of Clinical Child Psychology*, 15, 260-267.
- Centre for Reviews and Dissemination (2009). *Systematic reviews: CRD's guidance for undertaking reviews in healthcare*. York: CRD, University of York.
- Cohen, J. (1960). A coefficient of agreement for nominal scales. *Educational and Psychological Measurement*, 20, 37-46.
- Davis, H. (1993). *Counselling parents of children with chronic illness or disability*. Leicester: BPS Books.
- Davis, E., Shelly, A., Waters, E., Boyd, R., Cook, K., & Davern, M. (2009). The impact of caring for a child with cerebral palsy: quality of life for mothers and fathers. *Child: Care, Health and Development*, 36(1), 63-73.
- Deb, S. & Prasad, K. B. (1994). The prevalence of autistic disorder among children with a learning disability. *British Journal of Psychiatry*, 165, 395-399.
- Dunn, M. E., Burbine, T., Bowers, C. A., & Tantleff-Dunn, S. (2001). Moderators of stress in parents of children with autism. *Community Mental Health Journal*, 37, 39-52.

- Edwards, M., & Titman, P. (2010). *Promoting psychological well-being in children with acute and chronic illness*. London: Jessica Kingsley.
- Eiser, C. (1990). Psychological effects of chronic disease. *Journal of Child Psychology and Psychiatry*, 31, 85-98.
- Foster, R. H., Kozachek, S., Stern, M., & Elsea, S. H. (2010). Caring for the caregivers: an investigation of factors related to well-being among parents caring for a child with Smith-Magenis syndrome. *Journal of Genetic Counselling*, 19, 187-198.
- Fotiadou, M. Barlow, J H., Powell, L. A., & Langton, H. (2008). Optimism and psychological well-being among parents of children with cancer: An exploratory study. *Psycho-Oncology*, 17, 401-409.
- Friedrich, W. N., Wiltner, L. T., & Cohen, D. S. (1985). Coping resources and parenting mentally retarded children. *American Journal on Mental Retardation*, 90, 130-139.
- Friedrich, W. N., Cohen, D. S. & Wiltner, L. T. (1988). Specific beliefs as moderator variables in maternal coping with mental retardation. *Children's Health Care*, 17(1), 40-44.
- Glenn, S., Cunningham, C., Poole, H., Reeves, D., & Weindling, M. (2009). Maternal parenting stress and its correlates in families with a young child with with cerebral palsy. *Child: Care, Health and Development*, 35(1), 71-78.
- Goodley, D. & Tregaskis, C. (2006). Storying disability and impairment. Retrospective accounts of disabled family life. *Qualitative Health Research*, 16(5), 630-646.
- Goldberg, L. R. (1999). *A broad-bandwidth, public domain, personality inventory measuring the lower-level facets of several five-factor models*, in I. Mervielde, I. Deary, F. De Fruyt & F. Ostendorf (eds) *Personality Psychology in Europe*, vol. 7, pp. 7-28. Tilburg, The Netherlands: Tilburg Universities Press.
- Hagekull, B., Bohlin, G., & Hammarberg, A. (2001) The role of parental perceived control in child development: a longitudinal study. *International Journal of Behavioral Development*, 25, 429-437.
- Hamlyn-Wright, S. Draghi-Lorenz, R., & Ellis, J. (2007). Locus of control fails to mediate between stress and anxiety and depression in parents of children with a developmental disorder. *Autism*, 11(6), 489-501.
- Hassall, R., & Rose, J. (2005). Parental cognitions and adaptation to the demands of caring for a child with an intellectual disability: A review of the literature and implications for clinical interventions. *Behavioural and Cognitive Psychotherapy*, 33, 71-88.
- Hassall, R., Rose, J., & McDonald, J. (2005). Parenting stress in mothers of children with an intellectual disability: The effects of parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research*, 49, 405-418.
- Hastings, R. P., Beck, A., & Hill, C. (2005). Positive contributions made by children with an intellectual disability in the family: Mothers' and fathers' perceptions. *Journal of Intellectual Disabilities*, 9, 155-165.

- Hastings, R. P., Kovshoff, H., Brown, T., Ward, N. J., Espinosa, F.D., & Remington, B. (2005). Coping strategies in mothers and fathers of preschool and school-age children with autism. *Autism*, 9, 377-391.
- Henderson, D., & Vandenberg, B. (1992). Factors influencing adjustment in the families of autistic children. *Psychological Reports*, 71, 167-171.
- Henry, D.B., Tolan, P.H., & Gorman-Smith, D. (2005). Cluster analysis in family psychology research. *Journal of Family Psychology*, 19(1), 121-132.
- Hill R. (1949). *Families under stress*. New York, NY: Harper and Row.
- Hill R. (1958). Social stresses on the family. *Social Case Work*, 49, 139-150.
- International Association for the Scientific Study of Intellectual and Developmental Disabilities (2012). Position paper, July 2012: Families supporting a child with intellectual or developmental disabilities: The current state of knowledge. Retrieved July 29, 2012 from :
iassid.org/images/documents/SIRGs/Families/families_sirg_position_paper_final.pdf
- International Personality Item Pool. (2002). *A Scientific collaboratory for the development of advanced measures of personality traits and other individual differences*. Retrieved July 9, 2012 from: <http://ipip.ori.org>.
- James, W. H., & Rotter, J. B. (1958). Partial and 100% reinforcement under chance and skill conditions. *Journal of Experimental Psychology*, 55, 397-403.
- Janssens, J. M. A. M. (1994). Authoritarian child rearing, parental locus of control, and the child's behaviour style. *International Journal of Behavioral Development*, 17(3), 485-501.
- Jones, J., & Passey, J. (2005). Family adaptation, coping and resources: Parents of children with developmental disabilities and behaviour problems. *Journal on Developmental Disabilities*, 11, 31-46.
- Ketelaar, M., Volman, M. J. M., Gorter, J. W., & Vermeer, A. (2008). Stress in parents of children with cerebral palsy: What sources of stress are we talking about? *Child: Care, Health and Development*, 34(6), 825-829.
- Konstantareas, M. M., & Lampropoulou, V. (1998). Stress in Greek mothers with deaf children: Effects of child characteristics, family resources and cognitive set. *American Annals of the Deaf*, 140(3), 264-270.
- Kuster, P. A., & Badr, L. K. (2006). Mental health of mothers caring for ventilator-assisted children at home. *Issues in Mental Health Nursing*, 27, 817-835.
- Lanfranchi, S., & Vianello, R. (2012). Stress, locus of control and family cohesion and adaptability in parents of children with Down, Williams, Fragile X, and Prader-Willi syndromes. *American Journal on Intellectual and Developmental Disabilities*, 117(3), 207-224.
- Lazarus, R. S. (1991). *Emotion and adaptation*. New York, NY: Oxford University Press.
- Lazarus, R. S. & Folkman, S. (1984). *Stress, appraisal, and coping*. New York, NY: Springer.
- Lefcourt, H. M. (1982). *Locus of control: Current trends in theory and research*. Hillsdale, NJ: Lawrence Erlbaum Associates.

- Lloyd, T. & Hastings, R.P. (2009). Parental locus of control and psychological well-being in mothers of children with intellectual disability. *Journal of Intellectual and Developmental Disability*, 34(2), 104-115.
- Mash E. J. & Johnston C. (1990) Determinants of parenting stress: Illustrations from families of hyperactive children and physically abused children. *Journal of Clinical Child Psychology*, 19, 313-328.
- McCann, D., Bully, R., & Winzenberg, T. (2012). The daily patterns of time use for parents of children with complex needs: A systematic review. *Journal of Child Health Care*, 16(1), 26-52.
- McCubbin H. I. & Patterson J. M. (1983). The family stress process: The Double ABCX Model of adjustment and adaptation. In: H. I. McCubbin, M. B. Sussman & J. M. Patterson (eds), *Social Stress and the Family: Advances and Developments in Family Stress Theory and Research*, pp. 7-37. New York, NY: Haworth Press.
- McKinney, B., & Peterson, R. A. (1987). Predictors of stress in parents of developmentally disabled children. *Journal of Pediatric Psychology*, 12(1), 133-150.
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLoS Medicine*, 6(6). Retrieved 17 February 2012 from plosmedicine.org/article/info%3Adoi%2F10.1371%2Fjournal.pmed.1000097
- Neece, C. L., Green, S. A., & Baker, B. L. (2012). Parenting stress and child behavior problems: A transactional relationship across time. *American Journal on Intellectual and Developmental Disabilities*, 117(1), 48-66.
- Olsson, M. B., & Hwang, C. P. (2008). Socioeconomic and psychological variables as risk and protective factors for parental well-being in families of children with intellectual disabilities. *Journal of Intellectual Disability Research*, 52(12), 1102-1113.
- Phares, V., Fields, S., Kamboukos, D., & Lopez, E. (2005). Still looking for poppa. *American Psychologist*, 60, 735-736.
- Rimmerman, A. (1991). Parents of adolescents with severe intellectual disability in Israel: Resources, stress and the decision to apply for out-of-home placement. *Australia and New Zealand Journal of Developmental Disabilities*, 17, 321-329.
- Rotter, J. B. (1975). Some problems and misconceptions related to the construct of internal versus external control of reinforcement. *Journal of Consulting and Clinical Psychology*, 43, 56-67.
- Scottish Intercollegiate Guidelines Network (2008). *SIGN 50: A guideline developer's handbook*. Edinburgh: Author.
- Seligman, M. E. P. (1975). *Helplessness: On depression, development, and death*. San Francisco, CA: W. H. Freeman.
- Siman-Tov, A., & Kaniel, S. (2011). Stress and personal resource as predictors of the adjustment of parents to autistic children: A multivariate model. *Journal of Autism and Developmental Disorders*, 41, 879-890.

- Singer, G. H. S. (2006). Meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities. *American Journal on Mental Retardation*, 111(3), 155-169.
- Wallander, J. L., & Varni, J. W. (1992). Adjustment in children with chronic physical disorders: Programmatic research on a disability-stress- coping model. In A. M. La Greca, L. J. Siegel, J. L. Wallander, & C. E. Walker (Eds.), *Stress and coping in child health. Advances in pediatric psychology* (pp. 279-298). New York, NY: Guilford Press.
- Wallander, J. L., & Varni, J. W. (1998). Effects of pediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry*, 39(1), 29-46.
- Wallander, J. L., Varni, J. W., Babani, L., DeHaan, C. B., Wilcox, K. T., & Banis, H. T. (1989). The social environment and the adaptation of mothers of physically handicapped children. *Journal of Pediatric Psychology*, 14, 371-387.
- Weiss M. J. (2002). Hardiness and social support as predictors of stress in mothers of typical children, children with autism, and children with mental retardation. *Autism*, 6, 115-130.

CHAPTER 2: INTRODUCTION TO STUDY

This chapter briefly revisits the theoretical background and key findings of the systematic review in order to set the context and develop the rationale for the current study. The research hypotheses of the study are then subsequently outlined. The study will investigate whether cognitive factors predict parental subjective wellbeing in a specific group of parents who have children with profound and multiple intellectual disabilities (PMID). Children with PMID are a heterogeneous group who have a profound intellectual disability, in addition to multiple sensory, motor and other impairments. They also commonly have a multitude of medical conditions that require complex management and that can be life limiting, representing a number of challenges for family caregivers that will be explored in more detail in the next chapter.

Theoretical background

Four key theoretical models were outlined in the systematic review chapter: the Stress-Coping Model (Lazarus & Folkman, 1984), the Stress-Coping Model for Coping with Chronic Disease (Lazarus, 1991), the Double ABCX Model (McCubbin & Patterson, 1983) and the Parent Disability-Stress-Coping Model (Wallander *et al.*, 1989). All of these models reinforced the multi-dimensional and transactional nature of the process of responding to stress in various contexts. Cognitive, emotional and behavioural responses to a stressor are central variables that influence outcomes. These can be at the level of an individual, family, or wider system and are partly determined by the wider environment and available resources. Cognitive appraisals are particularly important, as they affect whether a situation is perceived as positive or negative, and within or outside one's control. This then influences an individual's or family's emotional and behavioural responses and determines the coping strategies they may or may not employ. Different coping styles are associated with different outcomes, thus cognitive appraisals have an important role in mediating the relationship between a stressor and outcome.

Locus of control

As outlined in the systematic review, locus of control is a specific type of cognitive appraisal involved in this process and the concept is derived from Rotter's (1954) social learning theory. This proposed that the potential occurrence of behaviours that satisfy some need is a function of the expectancy that those behaviours will lead to these reinforcements, in addition to the strength of value these reinforcements have. A key aspect of this is the individual's interpretation of the cause of their experiences. Rotter (1966) observed that some individuals adapt their behaviour following new experiences and gain from this, whilst others attribute change to others, rather than their own behaviour or characteristics. Expectancies regarding the controllability of outcomes thus determine behavioural responses and other cognitive appraisals (James & Rotter, 1958). Furthermore,

controllability judgements affect subsequent responses to that and other situations through a causal analysis of success and failure (Lefcourt, 1982).

Parental locus of control

Locus of control is deemed situation specific and can vary depending on the situation and the behaviour (Rotter, 1975). Parental locus of control relates to a parent's experience of parenting their child, specifically how much they perceive the child's behaviour and development are a result of the parenting they receive and how much control they have over these child outcomes as their parent (Lloyd & Hastings, 2009a). A parent with an internal parental locus of control will believe that they have some control over the child, their behaviour and their development. Whereas a parent with an external parental locus of control will perceive that they have little or no control over the child, their behaviour and their development.

Parental locus of control and parenting a child with a disability

The systematic review revealed a strong relationship between parental locus of control and several parental psychological outcomes. An external parental locus of control was linked with higher levels of stress (Hassall *et al.*, 2005; Lloyd & Hastings, 2009a; Siman-Tov & Kaniel, 2011), anxiety (Lloyd & Hastings, 2009a), and depression (Dunn *et al.*, 2001; Lloyd & Hastings, 2009a), in addition to use of more maladaptive coping strategies (Dunn *et al.*, 2001). An internal parental locus of control was associated with lower levels of parental stress (Dunn *et al.*, 2001), the ability to recognise positive gains of having a child with a disability (Lloyd & Hastings, 2009a) and increased family cohesion (Glenn *et al.*, 2009).

In addition, parental locus of control mediated the inverse relationship between stress levels of mothers of children with ID and family support in one of the studies examined in the systematic review (Hassall *et al.*, 2005). They suggested that family support facilitated the development of a more internal parental locus of control. Furthermore, social support has been found to indirectly affect parental adjustment and the reported severity of the child's symptoms via parental stress (Siman-Tov & Kaniel, 2011). Receiving support seems to lower stress by helping parents feel more in control and encouraging a sense of meaning (Siman-Tov & Kaniel, 2011). Therefore, parental locus of control influences parental psychological outcomes resulting from the experience of caring for a child with a disability, both directly and indirectly.

Other cognitive factors

In the context of parenting a child with a disability, parental locus of control is one of a number of cognitive factors associated with resilience in parents of children with ID. These comprise self-efficacy (Hastings & Brown, 2002), hope (Lloyd & Hastings, 2009b), optimism

(Baker *et al.*, 2005), benefit finding (Rapanaro *et al.*, 2008), acceptance (Lloyd & Hastings, 2008) and mindfulness (Singh *et al.*, 2006).

The ability to report positive experiences of parenting a child with additional needs has also been implicated in parent psychological outcomes (Glenn *et al.*, 2009; Hassall *et al.*, 2005; Lloyd & Hastings, 2009a). There is evidence that maternal positive perceptions of having a child with a disability are associated with reframing as a coping strategy (Hastings *et al.*, 2002). Parental beliefs are also significant in terms of parents' appraisal of the efficacy of behavioural interventions, with beliefs of intervention efficacy associated with lower parenting stress (Hastings & Johnson, 2001).

Positive psychological outcomes for parents who have a child with a disability

Psychology as a field has devoted more attention to unhappiness and dysfunction than to the determinants and consequences of positive functioning (Diener, 1984). The literature on factors that promote and facilitate positive psychological outcomes for parents of children with disabilities follows this pattern and is less developed than that investigating parenting stress in this group (Olsson & Hwang, 2008). It is important to look at families that do well, as well as families who struggle, given that parents report both positive and negative experiences related to having a child with a disability (Heiman, 2002; Lloyd & Hastings, 2009b). It could be argued that the existing literature does indeed investigate factors that promote wellbeing, as one of the sequelae of investigating stress. Nevertheless, there are problems with using an 'absence of' stress, distress or mental health issues as a proxy measure of wellbeing (Ruini *et al.*, 2003). Research incorporating both positive and negative psychological outcomes has revealed that they are independent of one another (Hastings & Taunt, 2002), thus the absence of one should not necessarily be interpreted as the presence of the other.

With the growth of wellbeing research and positive psychology more generally, there has been a proliferation of research explicitly investigating adaptive psychological functioning and positive psychological outcomes, such as subjective wellbeing, adjustment, quality of life, satisfaction, adaptation and acceptance (Seligman & Csikszentmihalyi, 2000). This has resulted in the development of a wide range of tools to measure these concepts, although none have been developed specifically for parents of children with a disability. However, positive psychology has been described as an area that is gaining credence in the field of intellectual disability research (Lloyd & Hastings, 2009b).

Further research is needed to identify factors that promote positive psychological outcomes such as parental wellbeing and adjustment to parenting a child with a disability. Explicitly investigating these, in addition to factors that protect against the negative impact of the stressors inherent in the experience of having a child with a disability, will add significantly

to the literature and provide a useful basis for developing interventions. The development or adaptation of psychometrically robust measures for this purpose will also facilitate and encourage further research in this area that explicitly focuses on aspects of positive psychological outcomes in this group of parents.

Rationale and aims for the current study

Further research that focuses on factors that promote positive psychological outcomes and adaptive psychological functioning of caregivers and families of children with disabilities will enable the development of an evidence base to inform interventions. Cognitive factors have been found to have both a direct and indirect role in determining psychological outcomes for this group and are one area where interventions could be developed, as cognitions can be amenable to change.

Consequently, one of the research aims for this study is to identify cognitive factors that predict subjective wellbeing in parents of children with a disability. Parental locus of control and positive perceptions of their child were selected as the two dependent variables to investigate in the current study, as the literature reviewed indicated that these both predict positive psychological outcomes in parents of children with a disability.

Rationale for chosen population

The current study will focus on a specific subgroup of family caregivers of children who have PMID. This group is relatively underrepresented in the research literature and thus little is known about the applicability of existing models of caregiving stress to this distinct subgroup of caregivers. Children with PMID have a very specific pattern of needs in terms of multiple physical disabilities, medical conditions and severely impaired cognitive functioning, which vary hugely between individuals. These are some of most vulnerable young people in society and there are multiple care tasks associated with supporting them.

It is important to specifically consider this group of caregivers as they are at increased risk of negative psychological outcomes due to manifesting multiple risk factors identified in the literature. These include caring for a child with chronic and potentially life-limiting physical health issues that often require complex medical management, who is both severely cognitively and functionally impaired and requires a high level of constant support. The emotional effects of this, such as feelings of guilt, loss, anxiety, depression and anger increase the likelihood of poor psychological functioning. The wider effects on caregivers and their families' lives in terms of reducing the likelihood of protective factors such as social supports, a career, and healthy relationships with significant others also mean that this is a group particularly at risk of poor psychological outcomes. The broader impact of this is also important to consider, in terms of the financial, community and societal costs associated with caregiver and family strain.

Hypotheses

This study will investigate whether two types of parental cognitions, parental locus of control and positive perceptions of their child, predict the subjective wellbeing of parents and family caregivers of children with PMID. It will also investigate whether any of these factors are significantly related to one another. The subjective wellbeing of this group will also be compared to that reported in the general population and in parents of children with behavioural difficulties who do not have a disability.

It is hypothesised that:

- (i) Self-reported parental locus of control will be predictive of self-reported parental subjective wellbeing.
- (ii) Self-reported recognition of the positive gains of having a child with PMID will be predictive of self-reported parental subjective wellbeing.
- (iii) Self-reported subjective wellbeing will be lower in parents and family caregivers of children and young people with PMID than in parents of children with behavioural difficulties who do not have a disability, and the general population.

CHAPTER 3: JOURNAL ARTICLE

This chapter summarises the background, aims, method, results and findings of the research study in the format of a journal article. A discussion of implications of these findings with regard to clinical practice and future research also follows, in addition to the references for the journal article. The style guidelines of the journal *Research in Developmental Disabilities* are adopted.

JOURNAL ARTICLE ABSTRACT

Parental locus of control predicts subjective wellbeing in parents who have a child with profound and multiple intellectual disability

Background: Parenting a child with a disability is associated with higher levels of stress, anxiety, and depression than parenting a typically developing child. However, some parents demonstrate considerable resilience and are able to adjust to this demanding role.

Aims: This study aimed to explore levels of parental subjective wellbeing in a specific group of these parents: those who have a child with profound and multiple intellectual disabilities (PMID). It also aimed to determine whether two types of parental cognition, parental locus of control and realisation of positive gains of having a child with PMID, were predictive of parental subjective wellbeing.

Method: A single sample of parents and family caregivers (N=101) completed three quantitative self-report questionnaires as part of a within-participant, cross-sectional survey design. These included the Positive Gain Scale, a modified version of the Parental Locus of Control Scale and the Warwick-Edinburgh Mental Wellbeing Scale.

Results: The median subjective wellbeing score for this group of parents was well below the equivalent value for the general population. Regression analysis revealed that parental locus of control significantly predicted parental subjective wellbeing ($\beta = -.279$, $t(2,99) = 9.419$, $p = .005$), accounting for around 8% of the variance in WEMWBS scores, adjusted $R^2 = .081$, $F(2,99) = 5.474$, $p = .006$.

Conclusions: These results highlight the importance of parental locus of control in influencing the subjective wellbeing of parents of children with PMID. This suggests a role for psychological intervention for parents and families with a focus on promoting an internal parental locus of control. However, further research is needed.

Highlights

- 101 parents of children with profound and multiple disability participated
- A single sample, cross-sectional survey design was employed
- Parental locus of control predicted parent subjective wellbeing
- Realisation of positive gains did not predict parent subjective wellbeing
- Further research is needed in this area

Keywords: intellectual, developmental, disability, parent, cognition, wellbeing

1. Introduction

1.1 Background

Around 2.5% of the UK population has an intellectual disability (ID; Michael, 2008), defined as significant impairment in the areas of cognitive and adaptive functioning that manifests before the age of 18 years (British Psychological Society, 2000). People with profound and multiple intellectual disabilities (PMID) are a sub-group of this population who need the highest levels of support with most aspects of daily life due to having more than one pervasive disability in several areas of functioning (Mencap & PMLD Network, n.d.). These include significant cognitive, sensory, communication and mobility impairments and complex physical and mental health needs (Mencap & PMLD Network n.d.).

1.2 Prevalence

There is a paucity of accurate information regarding the numbers of people with PMID, which is due in part to the differing criteria used to define this population (Hogg, 1999; Nakken & Vlaskamp, 2007; Pawlyn & Carnaby, 2009). Emerson and Hatton (2009) recently estimated that there were 12567 children and young people under 18 in England with PMID and highlight that the 'true' overall number of people with ID known to services is likely to increase in the UK by twenty per-cent over the period 2001-2021. The numbers of people with PMID are therefore likely to continue to increase significantly, due mainly to advances in medical care and improved support in developed countries such as the UK (Nakken & Vlaskamp, 2007).

1.3 Aetiology

The aetiology of PMID is highly variable and not always identifiable (Hogg, 1999), although it is accepted that sixty to seventy five per cent of biological causes occur prenatally (Pawlyn & Carnaby, 2009) and that significant neurological damage leads to a wide range of developmental difficulties compounded by physical health problems (Hogg, 1999). Genetic conditions such as Rett syndrome, Cornelia de Lange syndrome and Rubenstein-Taybi syndrome are associated with PMID (Arvio & Sillanpää, 2003) and cerebral palsy is the most common non-genetic cause (Pawlyn & Carnaby, 2009). Other causes include birth trauma and cerebral hypoxia perinatally, and accidents and infections such as meningitis, postnatally (Pawlyn & Carnaby, 2009). Psychosocial factors are not thought to be a major factor contributing to the occurrence of severe and profound intellectual disabilities (Emerson, Hatton, Felce, & Murphy, 2001).

1.4 Care needs

The physical health needs of this group are highly complex (Nakken & Vlaskamp, 2007), usually require 24 hour care and often include severe epilepsy, chronic respiratory difficulties, gastro-intestinal difficulties, sleep difficulties and dysphagia, among others

(Espie *et al.*, 1998; Hogg, 1999). The move from institution-based care of children with complex medical needs to increasingly home-based care in recent decades means that parents and caregivers often have to become highly skilled in terms of treatment regimes and medical equipment (Hatzmann Maurice-Stam, Heymans, & Grootenhuis, 2009). This can be extremely demanding for family caregivers and often impacts on other areas of their life, such as employment (Edwards & Titman, 2010).

Individuals with PMID are particularly vulnerable to illness and mortality due to the nature of their complex needs, although advances in medical technology have contributed to changes in patterns of morbidity and mortality (Department of Health, 2001; Kirk, 1998; Nakken & Vlaskamp, 2007). Severe problems with eating, drinking, sleeping, motor skills, toileting, self-care, emotion-regulation and communication are highly prevalent within this group as a consequence of their complex healthcare and psychological needs (Nakken & Vlaskamp, 2007). Sensory impairment and sensitivities are also an issue for people with PMID (Nakken & Vlaskamp, 2007). Consequently, individuals with PMID must completely rely on those around them to recognise and meet their complex needs accordingly (Nakken & Vlaskamp, 2007).

1.5 Psychological needs

As well as fragile physical health, individuals with PMID can also experience significant stress and anxiety as a result of having very limited control over their environment (Hogg, 1999). There is evidence that they may experience distress, even when no overt signs of this are present (Chaney, 1996). People with PMID are also very sensitive to the emotional state of mind of those caring for them, in addition to commotion, co-operation and tension in the environment in which they are being cared for (Pettry *et al.*, 2007). They also have difficulties understanding and communicating their needs and rely on those around them to recognise and interpret often idiosyncratic patterns of behaviour, which can be a complex and challenging process for caregivers (Bunning, 2009). Family and frontline caregivers are usually the most sensitive to changes in the person with PMID that can indicate psychological distress, but may not have the knowledge or skills to recognise and prevent difficulties (Moss, Bouras, & Holt, 2000). It has been reported that they are also often unaware that the person they are caring for can be referred for psychological or psychiatric assessment and intervention (Hogg, 1999). Parents and other caregivers have anecdotally reported anxiety and depression in those they care for with PMID (Nind, 2009), but in both research and clinical practice, accurate and reliable identification of mental health problems in this group is difficult (*cf.* Hatton, 2002).

1.6 Impact on caregivers

In addition to the emotional distress experienced by people with PMID, their behaviour may sometimes be challenging and distressing to those around them. This might include the

production of sounds or noises that are disruptive or difficult to ignore (e.g. crying, screaming), ingestion of inappropriate objects or substances, and in particular, self-injurious behaviour (Hogg & Lambe, 1988). This can be very distressing for caregivers to witness and difficult and unsafe to ignore, thus making these behaviours challenging and stressful to manage (Bunning, 2009). It has been widely acknowledged that parents' commitment to caring for a child with PMID is unconditional and long-term (Hogg & Lambe, 1988). However, this caring role presents a set of demands that are somewhat different to caring for more able relatives or those with fewer health issues (Hogg, 1999). Caring for someone with such a range of pervasive, complex needs that often follow an unpredictable course, results in an extensive variety and range of caring activities (Raina *et al.*, 2004). All of these factors represent a number of significant stressors inherent in this caring role (Lambe, 1998), which for family caregivers is often not optional.

The burden of care experienced by this group of caregivers affects their quality of life and puts them at particular risk of experiencing adverse psychological consequences (Hatzmann *et al.*, 2009). It is well-established that parents of children with significant illness or disability are more likely to experience stress, anxiety, depression and decreased wellbeing than parents of children who do not have these difficulties (Foster, Kozachek, Stern & Elsea, 2010; Fotiadou, Barlow, Powell, & Langton, 2008; Ketelaar, Volman, Gorter & Vermeer, 2008; Kuster & Badr, 2006; Neece, Green & Baker, 2012; Singer, 2006). Greater caring demands and higher levels of motor and cognitive impairment in those being cared for have been associated with higher levels of caregiver stress and burden (Deater-Deckhard, 2004; O'Neil, Palisano, & Wescott, 2001; Ong, Chandran, & Peng, 1999). Added to the more practical stressors are the emotional challenges of caring for a child with multiple disabilities and complex health issues that can be life limiting (Edwards & Titman; Fitton, 1994; Grove, 2007). These may include feelings of emotional pain, loss, guilt, anger, anxiety, helplessness, depression, as well as a range (and often mixture) of other emotions (Aldridge, 2007; Beckman, 1991). Furthermore, families exist as systems and are present within a wider context, thus the influence of external stressors not directly related to the person with PMID can also add extra pressures to family caregivers and wider networks (Raina *et al.*, 2004; Wallander & Varni, 1998).

1.7 Caregiver wellbeing

Parents report both positive and negative experiences related to having a child with a disability (Lloyd & Hastings, 2009a). Therefore, positive experiences of caregivers need to be investigated in order to understand factors associated with caregiver wellbeing and adaptation, as well as the stressors and negative outcomes associated with this role (Kearney & Griffin, 2001; Olsson & Hwang, 2008). There has been a renewed interest in wellbeing research and positive psychology in recent years, resulting in a proliferation of research focusing on adaptive psychological functioning and positive psychological

outcomes, such as subjective wellbeing, adjustment, quality of life, satisfaction, adaptation and acceptance (Seligman & Csikszentmihalyi, 2000). Positive psychology has been described as an area that is gaining credence in the field of ID research (Lloyd & Hastings, 2009a) and recent studies have investigated hope (e.g. Lloyd & Hastings, 2009a), wellbeing (e.g. Glidden, Billings, & Jobe, 2006), optimism (e.g. Baker, Blacher, & Olssen, 2005), and acceptance (Lloyd & Hastings, 2008) in parents of children with ID. A better understanding of factors that facilitate subjective wellbeing and adaptation in parents who have a child with PMID will provide a theoretical basis for further research with this population, in addition to informing the development of interventions and service provision to support this group of parents.

2. Aims and hypotheses

This study aimed to explore the subjective wellbeing of parents and family caregivers of children and young people with PMID, in relation to comparisons of subjective wellbeing reported in the general population, and the influence of parental cognitive factors¹.

It was hypothesised that:

- (i) Self-reported parental locus of control would be predictive of self-reported parental subjective wellbeing.
- (ii) Self-reported recognition of the positive gains of having a child with PMID would be predictive of self-reported parental subjective wellbeing.
- (iii) Self-reported subjective wellbeing would be lower in parents and family caregivers of children and young people with PMID than in parents of children with behavioural difficulties who did not have a disability, and the general population.

3. Method

3.1 Participants

One hundred and one parents and family carers with at least one child with PMID (97 females, 4 males; age range 20-70 years) participated in this cross-sectional, self-report questionnaire-based survey. Given the self-selecting nature of participation, those who took part were people who viewed themselves as a parent or carer. The term 'carer' was purposefully not defined, given the multitude of different conceptualisations of this role (Bytheway & Johnson, 2008) and in order to capture data from a broad range of individuals in a caring role that was not their employment (with the exception of foster carers).

¹ Based on power calculations, a pragmatic decision was taken to limit the number of variables investigated to two predictor variables and one criterion variable. This decision was based on the potential difficulty identified in accessing the study population and expectations of recruitment difficulties due to the nature of participants' caring demands.

This study adopted the criteria published in Mencap and the PMLD Network's leaflet on the definition of PMID (n.d.). These included children and young people who needed high levels of support with most aspects of daily life due to having more than one disability, including:

- (i) a profound intellectual disability*
- (ii) significant communication difficulties*
- (iii) sensory impairment (e.g. hearing, vision)*
- (iv) physical disabilities and mobility problems*
- (v) complex health needs (e.g. enteral/parenteral feeding, ventilation/CPAP)*
- (vi) mental health difficulties*

Demographic characteristics of the caregivers who participated and the children they cared for are summarised in Table 1. Caregivers in this study were predominantly white (97%), female (96%), a parent of the child (96%), and married, in a civil partnership or cohabiting (71%). Most caregivers were aged 41-50 years (40%), followed by the 31-40 age group (32%). In terms of socio-economic status, the majority were employed or in education (52%), educated to undergraduate level (30%), and owner-occupiers of their homes (67%). Almost half of the caregivers surveyed (45%) reported that they had their own health issues (e.g. anxiety, back pain, cancer, depression, fibromyalgia, high blood pressure, irritable bowel syndrome, lupus, osteoarthritis, previous stroke and psoriasis).

Twenty per cent of children with PMID were living in a one-parent household. At least 63 per cent of the children with PMID had siblings living in the same family home, although 13 per cent of the sample did not provide information for this item. Thirty-seven per cent of the overall sample had one sibling at home, nineteen per cent had two siblings at home and eight per cent had three or more siblings at home. Children with PMID had a range of conditions, including genetic and chromosomal abnormalities (e.g. Cri du Chat syndrome, Phelan McDermid syndrome and Di George syndrome), cerebral palsy, epilepsy, scoliosis, gastrointestinal issues and hearing or vision impairment.

Participants were recruited via two routes, including two local independent schools with special provision for children and young people with PMID. Recruitment was also undertaken via parent support groups, voluntary organisations and charities for children with disabilities, physical health conditions (including genetic conditions, chromosomal abnormalities and syndromes) and/or complex needs.

Table 1: Summary of caregiver and child demographic characteristics

		% of sample, N= 101
Age in years	≤ 20	1
	21-30	8
	31-40	32
	41-50	41
	51-60	18
Gender	Male	4
	Female	96
Relationship to child with PMID	Parent	96
	Foster carer	2
	Other	3
Ethnicity	White	97
	Other	3
Employed/studying		51
Married/civil partnership/cohabiting		79
Own health issues		45
One parent household		20
n= 78		
Siblings at home	1	37
	2	19
	≥ 3	8
Child with PMID gender	Male	60
	Female	40
Child with PMID age in years	Mean	10.33
	SD	5.85
	Range	1-23
n= 88		

3.2 Ethical considerations

Ethical approval was granted by the University of Edinburgh School of Health in Social Science Ethics Committee. The potentially distressing nature of the study was highlighted in the participant information sheet prior to participation. Information was also provided about how participants could seek further support. Participants' right to withdraw was highlighted at several stages.

3.3 Procedure

Information was provided to parents and caregivers regarding the study via electronic or paper participant information sheets. Participation was voluntary and it was made clear that whether or not they took part would not affect the services families received in any way. Completion and submission of the questionnaires was deemed as giving informed consent for responses to be used for the purpose of the study and participants were made aware of this. The survey was made available both as an anonymous hard copy paper booklet and also as an online survey accessed via a secure website. Participants were first asked to provide demographic information about themselves, their child and anyone else living within the home. Three self-report questionnaires were then presented and space provided for any feedback parents wished to provide.

3.4 Self-report questionnaires

3.4.1 Parental Locus of Control Scale - Revised Version

The Parental Locus of Control Scale - revised version (PLOC-R; Lloyd & Hastings, 2009) was used to measure parents' locus of control within the context of the parent-child relationship. The scale has been adapted specifically for parents of children with ID (*cf.* Campis, Lyman, & Prentice-Dunn, 1986 for original version). The revised scale includes 42 statements that parents respond to in terms of how much they agree or not, using a four-point Likert-type scale. Subscales include items that explore parental efficacy, responsibility, belief in fate/chance and child control over parents' life. For the purpose of this study, this was changed to a five-point likert scale by adding 'not sure' as a mid-point response, in line with the other questionnaires used in the study. Using the five-point scale, the internal consistency of the PLOC-R was good, with a Cronbach's alpha (Cronbach, 1951) of .92. A PLOC-R total score was used as a predictor variable to investigate the relationship between parental locus of control and parental subjective wellbeing. The maximum score on this scale is 210, with a high score indicating an external locus of control in relation to the respondent's parenting of their child.

3.4.2 Positive Gain Scale

The Positive Gain Scale (PGS; Pit-ten Cate, 2003) was used to measure positive experiences associated with raising a child with disability. The measure consists of seven items; five relating to the perceived benefits for the parent of raising a child with a disability and two focusing on what the family has gained. Responses to each of the seven statements are given using a five-item Likert type scale, ranging from 'strongly agree' to 'strongly disagree'. Preliminary research findings have indicated that the PGS has face and content validity for parents of children with hydrocephalus and spina bifida (Pit-ten Cate, 2003). For the current study, the PGS demonstrated an acceptable level of internal consistency, with a Cronbach's alpha (Cronbach, 1951) of .70. A PGS total score was used as a predictor

variable in the current study to investigate the relationship between parents' positive perceptions of their child with PMID and parental subjective wellbeing. The maximum score on this scale is 35, with a low score indicating higher positive gains reported by parents.

3.4.3 Warwick-Edinburgh Mental Wellbeing Scale

The Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS; Stewart-Brown *et al.*, 2009; Tennant, Fishwick, Platt, Joseph, & Stewart-Brown, 2007) was used to measure parents' subjective psychological wellbeing. The Warwick-Edinburgh Mental Well-being Scale was funded by the Scottish Government National Programme for Improving Mental Health and Well-being, commissioned by NHS Health Scotland, developed by the University of Warwick and the University of Edinburgh, and is jointly owned by NHS Health Scotland, the University of Warwick and the University of Edinburgh. It is a brief, 14 item scale covering most aspects of positive mental health (positive thoughts and feelings) and psychological wellbeing currently in the literature, including both hedonic and eudaimonic perspectives. All items are worded positively and address aspects of positive mental health. Responses to each of the 14 statements are given using a five-item Likert type scale, ranging from 'none of the time' to 'all of the time'. The scale has been validated for use in the UK with those aged 16 and above and was standardised with both student and general population samples, in addition to focus group exploration of acceptability. The WEMWBS has been found to be psychometrically robust in terms of principal components factor analysis, construct validity, internal consistency, test-retest reliability, response bias, face/content validity, and Rasch analysis (Tennant *et al.*, 2007). For the current study, internal consistency was high, with a Cronbach's alpha (Cronbach, 1951) of .87.

As there is no specific measure of subjective wellbeing specifically designed for parents of children with ID, the WEMWBS was selected due to its general nature. The measure has been used to assess parental wellbeing in an evaluation of parenting interventions (Lindsay *et al.*, 2008). It was also selected on the basis of the measure being positively focused on wellbeing as opposed to problem-oriented. A WEMWBS total score was used to explore relationships between positive child perceptions, locus of control and parental subjective wellbeing. The maximum score on this scale is 70, with higher scores representing higher subjective psychological wellbeing.

4. Results

Summary statistics for each of the three key variables are provided in Table 2.

Table 2: Key variable summary statistics

	Mean	SD	Median	Range
Parental locus of control <i>PLOC-R total score</i>	113.42	16.47	115.00	66-153
Positive gains <i>PGS total score</i>	23.51	8.56	26.00	1-35
Parental subjective wellbeing <i>WEMWBS total score</i>	39.49	7.57	39.00	22-60

4.1 Relationships among key variables

The only statistically significant relationship to emerge among the key variables was a significant negative correlation between PLOC-R total scores and WEMWBS total score, $r = -.295$, $p = .003$. This means that higher scores on the PLOC-R (indicating an external parental locus of control) were associated with lower parental subjective wellbeing scores, which related to lower psychological wellbeing. There were no statistically significant relationships observed between PLOC-R total scores and PGS transformed total scores, nor between PGS transformed total scores and WEMWBS total scores.

4.2 Relationships with other variables

There were no statistically significant relationships observed between child age and PLOC-R total score, WEMWBS total score and PGS transformed total score. Statistically significant associations were revealed, however, between parental health status (two categories: identified health issues or no identified health issues) and both child age ($r = .213$, $p = .047$), and the PGS transformed total scores ($r = .604$, $p < .001$).

4.3 Subjective wellbeing of parents of children with PMID

The median WEMWBS total score of this sample was 39, more than 10 points below the general population median of 51 reported by Tennant *et al.* (2007) and less than the median of 43 reported for a group of parents of children who did not have a disability taking part in an early intervention project (Lindsay *et al.*, 2008).

4.4 Role of parental cognitions in predicting parental subjective wellbeing

Data relating to parents' total scores on the PLOC-R and transformed data relating to total scores on the PGS were entered into a multiple linear regression analysis (enter method) as a block of predictor variables at Step 1. Logarithmic transformation was applied to the PGS

total scores prior to this along with a constant (-1), in order to enable parametric analyses. The regression analysis aimed to determine whether the two types of parental cognition could predict parental subjective wellbeing and were entered this way as the analyses were exploratory (Brace *et al.*, 2009). Parental locus of control significantly predicted parental subjective wellbeing, $B = -.279$, $t(2,99) = 9.419$, $p = .005$. Parental locus of control explained around eight per cent of the variance in WEMWBS scores, adjusted $R^2 = .081$, $F(2,99) = 5.474$, $p = .006$. However, the regression analysis also revealed that realisation of the positive gains of having a child with PMID did not significantly predict parental subjective wellbeing and did not account for any of the variance in WEMWBS scores.

Table 3: Multiple regression analysis to predict parental subjective wellbeing

Variables	<i>B</i>	<i>SE B</i>	<i>B</i>	<i>t</i>	<i>p</i>
Step 0					
Constant	59.521	6.319		9.419	<0.001
Step 1					
PLOC-R	-0.131	0.045	-0.279	-2.895	0.005
PGS-LT	-1.977	1.649	-0.115	-1.199	0.233

B = unstandardised beta coefficient, *SE B* = standard error,
B = standardised beta coefficient, *t* = test statistic, *p* = significance value
PLOC-R = Parental Locus of Control Scale - Revised, PGS-T = Positive Gain Scale - log transformed data

5. Discussion

This study aimed to explore the subjective wellbeing of parents and family caregivers of children and young people with PMID, in relation to comparisons of subjective wellbeing reported in the general population, and the influence of parental cognitive factors.

5.1 Relationship between parental locus of control subjective psychological wellbeing

It was hypothesised that self-reported parental locus of control would predict self-reported parental subjective wellbeing in the sample of caregivers of children with PMID surveyed. A statistically significant inverse relationship between parental locus of control (measured using PLOC-R total scores) and parental subjective wellbeing (measured using WEMWBS total scores) emerged, with a medium effect size ($r = -.294$, $p = .003$). In this group of caregivers of children with PMID, a more external parental locus of control was related to lower levels of parental subjective wellbeing.

Furthermore, as hypothesised, multiple regression analysis revealed that parental locus of control significantly predicted subjective wellbeing of parents of children with PMID ($B = -$

.279, $t(2,99) = 9.419$, $p = .005$). Parental locus of control as a predictor accounted for around eight per cent of the variance in WEMWBS scores, $R^2 = .081$, $F(2,99) = 5.474$, $p = .006$. Both of these findings are in line with previous research highlighting a relationship between parental locus of control and negative parent psychological outcomes (Dunn *et al.*, 2001; Glenn *et al.*, 2009; Hassall *et al.*, 2005; Lloyd & Hastings, 2009;). These findings also fit with theoretical models that conceptualise the stress-coping process and aim to explain individual variation in parental outcomes (Lazarus & Folkman, 1984; Lazarus, 1991; McCubbin & Patterson, 1983; Wallander *et al.*, 1989).

5.2 Relationship between Recognition of positive gains of having a child with PMID and subjective psychological wellbeing

The current study found no statistically significant relationship between recognition of positive gains of having a child with PMID and parent subjective wellbeing. This was contrary to the findings expected and suggests that in this sample, being able to recognise positive aspects of having a child with PMID did not influence wellbeing, as has been observed in other studies involving more heterogeneous groups of parents of children with ID (e.g. Lloyd & Hastings, 2009a; Rapanaro *et al.*, 2008).

The distribution of caregiver PGS total scores revealed a pile-up of scores on the left hand side of the distribution that differed significantly from a normal distribution ($W = .909$, $df = 102$, $p < .001$). Caregiver total scores covered the full range that can be achieved on the seven item measure (1-35), suggesting no extreme floor effects were demonstrated. However, as a group, these caregivers achieved a relatively high mean total score of 23.51 ($SD = 8.56$) and the median value was similar at 26. Higher scores on this measure indicate recognition of fewer positive gains, thus there were some parents in this sample that found it difficult to see positive aspects of having a child with PMID.

Another possible explanation for these results is that caregivers were exhibiting a tendency to endorse statements that would be socially valued (e.g. 'Since having this child, my family has become closer to one another'), also known as the social desirability effect. Robinson and Anderson (1983) caution against reliance on only self-report measures in studies of parenting and family functioning for this reason. Indeed, given the fact that caregivers of children with PMID seldom seem to be asked how they feel about the experience of having a child with such complex needs (Fitton, 1994) and the paucity of research investigating their experiences, it is plausible that they may feel uncomfortable expressing thoughts that may not be socially desirable.

It must also be noted that the PGS only has seven items, whereas the other measures employed in the current study consisted of at least twice as many items. It may be that this measure does not include enough items to be sensitive enough to differences between

individuals within a group of parents who likely all value and see at least some positives in having such special children.

5.3 Influence of child age and parental health status

Although recognition of positive gains was not associated with parental subjective wellbeing or parental locus of control, a statistically significant relationship was revealed between transformed PGS total scores and whether the parent had identified that they had a health issue or not ($r = .604, p < .001$). This meant that parents who identified themselves as having a health issue were more likely to achieve higher scores on the PGS, indicating recognition of fewer positive gains. Parents with health issues may find it more difficult to see the positive gains of having a child with PMID because it is likely to impact negatively on their physical and mental health, although we cannot infer the direction of causality.

Furthermore, a statistically significant association between parental health status and child age was also revealed ($r = .213, p = .047$), indicating that parents with older children were more likely to be in the group with health issues. This would make sense in terms of the cumulative burden of the physical and emotional care demands associated with parenting a child with PMID needs (Nakken & Vlaskamp, 2007). However, child age was not significantly associated with any of the other variables investigated.

5.4 Subjective wellbeing of parents with PMID compared with other groups

As hypothesised, self-reported subjective wellbeing was lower in parents and family caregivers of children and young people with PMID than in parents of children with behavioural difficulties who did not have a disability, and the general population. The mean subjective wellbeing values of the sample studied ($M = 39.39; SD = 7.57$) corresponded with the 11th percentile for subjective wellbeing in the general population (Bryson *et al.*, 2011). This places parents of children with PMID alongside other groups experiencing low levels of support, autonomy, security and control in their lives or who are unemployed or experiencing levels of 'very bad' self-rated health (Bryson *et al.*, 2011). As well as subjective wellbeing being low in this group of caregivers in comparison with the general population, it was the equivalent of approximately the 25th percentile for pre-intervention subjective wellbeing in a group of parents taking part in a parenting early intervention programme (Lindsay *et al.*, 2008).

5.5 Clinical implications

The findings of this research study have implications for clinical practice, as practitioners often work closely with family members and caregivers as part of support provision for a child with PMID. The effect of parental locus of control on subjective wellbeing shows that it is how parents perceive and make sense of their situation that is key. Practitioner beliefs

may be very different to those of caregivers', thus practitioners need to remember not to make assumptions based on their own attributions.

The significant relationship between parental locus of control and caregiver subjective wellbeing observed suggests that it may be worth screening parents and other caregivers using brief measures of wellbeing, distress, parental locus of control or parental confidence. This would enable early identification of families who are likely to struggle, thus providing an opportunity for intervention and support before difficulties become salient at crisis. However, this clearly relies on the development and validation of psychometrically robust measures, whereas there are few available at the present time for parents of children with disabilities.

Following on from the identification of caregiver psychological needs, a range of interventions need to be developed specifically for this group of parents. Provision of realistic advice and guidance at an appropriate time for the family seems likely to have an impact on parental cognitive appraisals, such as locus of control. Cognitive appraisals are clearly important, thus existing interventions with a cognitive component are likely to be of some benefit to some caregivers, depending on their individual and family circumstances and beliefs. Social support in a formal group setting from peers in similar situations may be a powerful way of normalising caregivers' ideas and it also gives families the chance to check out some of their beliefs with other caregivers who understand the experience of having a child with PMID.

At present, the evidence base for interventions for parents of children with ID is almost non-existent. Some researchers in this area are advocating the use of acceptance and mindfulness based interventions for families of children with ID (MacDonald *et al.*, 2010). Interventions to help parents feel more empowered, capable and in control of parenting their child should be encouraged in whatever way possible. Practice-based research to evaluate any interventions that clinicians have found fruitful would be a good start to generating such an evidence base.

5.6 Limitations of the present study

The current study has a number of limitations that limit the generalisability of the findings. The number of variables investigated was limited to two predictors following power calculations, due to concerns regarding recruitment of this population to the study. Inclusion of a measure of child behaviour difficulties would have been particularly useful as this may have been related to parental locus of control and parental psychological wellbeing. An independent measure of child behaviour (i.e. not parent rated, as this would be influenced by parental perceptions of the behaviour) would have enabled consideration of a key stressor characteristic that is implicated in the literature and theoretical models

(Friedrich *et al.*, 1988; Hassall *et al.*, 2005; Konstantareas & Lapropoulou, 1995; McKinney & Peterson, 1987). Comparison of parent and independent ratings of child behaviour would also have been informative, as this may have elucidated the relationship between ‘actual’ stressor characteristics and ‘perceived’ stressor characteristics, referred to as primary and secondary appraisals in the Lazarus and Folkman (1984) stress-coping model.

The definition of PMID adopted in the current study was selected on the basis of it capturing the multiple physical disabilities and also the profound ID of these children, in combination with their fragile health and their functioning at the very early stages of development (Pawlyn & Carnaby, 2009). Caregivers were self-selected based on the definition of PMID provided and it was not possible to verify whether their child met these criteria or not. However, this is a common selection method in ID research (Emerson *et al.*, 2006) and given the wider issues of defining this group in research and practice, there were no obvious alternatives. Nakken and Vlaskamp (2007) highlight that this group has such profound ID that existing standardised tools for assessing intellectual functioning cannot offer a valid estimation of their intellectual capacity. Thus, there is a real need for assessment tools that are sensitive enough to confirm or disconfirm such impaired levels of functioning and that would both inform practice and enable clearer delineation of this population from other similar populations in research (Nakken & Vlaskamp, 2007).

Although efforts were made to encourage participation of fathers in this population, they still made up a very small proportion (3%) of the caregiver sample studied. An additional feature of this sample that limits generalisability is the fact that ninety-seven per cent of participants and ninety-four per cent of their children with PMID were of white ethnicity. There is evidence that families with a child with ID from minority ethnic groups consistently face disadvantages (e.g. financial) when compared with families who have a child with ID from majority ethnic communities and when compared to others from the same ethnic group (Hatton, 2002). It is likely that as the sample studied included a high proportion of caregivers who were white, the current findings may not represent the experiences of the wider population, including families from other ethnic backgrounds.

The cross-sectional design of the study limited the ability to explore causal and temporal relationships, although this is a limitation of much of the research on parental locus of control in this and similar populations (Siman-Tov & Kaniel, 2011). The discriminant validity of the Parental Locus of Control Scale requires further investigation, as it could be measuring other constructs such as parental distress or other cognitive variables (Lloyd & Hastings, 2009). However, this measure was selected for use in the current study as there is currently no psychometrically sound instrument available to measure parental locus of control that is developed specifically for, or is well validated with, caregivers of children with intellectual disability.

The current study focused on parental cognitions, but it is crucial to assess these findings within the wider context of family demographic and environmental factors (Olsson & Hwang, 2008). Emerson *et al.* (2006) urged researchers to consider the wider social context that families operate within, given that families of a child with a disability are at a greater risk of exposure to socio-economic adversity and that wellbeing and psychological distress are associated with social position (Power, 2002). Sufficient time and resources were not available to explore the full range factors that have been identified in the theoretical models of the stress-coping process. Therefore, the study focused on two specific cognitive factors identified as pertinent in the systematic review and wider literature. Clearly, the issues influencing subjective wellbeing in this group are complex and go beyond cognitive factors and further research is needed to test the various theoretical models outlined in full with this population of parents.

5.7 Directions for future research

It is likely that there is variation in child characteristics (e.g. child's current health status/prognosis, child behaviours, temperament) within this group of families, so it would be useful to investigate whether these are related to caregiver subjective wellbeing. In addition to this, it would be worth further exploring the association revealed in this study between caregiver characteristics (in this case health status) and both child characteristics and subjective wellbeing. It would also be useful to investigate whether locus of control influences subjective wellbeing and other psychological outcomes in paid caregivers and other staff working with children with PMID in caring roles.

The conceptual overlap between parental locus of control and related concepts such as parental self-efficacy, parent self-esteem, perceived competence and parental sense of coherence needs to be investigated further in order to delineate the contribution of each and further distinguish these from one another so that they represent useful concepts for research and practice.

The importance of relational variables such as relationship quality, attachment styles and social support were not investigated in this study. Future research exploring whether these relational variables are associated with parental cognitions would be valuable to our understanding of stress and coping within this group, particularly with regard to the identification of protective factors.

There is also a need for qualitative explorations of the experience of parents of children with PMID in order to gain an insight into their lived experiences and the emotional impact of caring for a very fragile child. Qualitative research could also explore caregivers' opinions on what they believe would be helpful or useful in relation to their own wellbeing.

5.8 Summary and conclusions

The present study provided further support for the relationship between parental locus of control and parental subjective wellbeing. Low levels of subjective wellbeing were observed in this group and the findings mirror previous research highlighting a relationship between parental locus of control and negative parent psychological outcomes (Dunn *et al.*, 2001; Glenn *et al.*, 2009; Hassall *et al.*, 2005; Lloyd & Hastings, 2009). Furthermore, this is the first known study to investigate the relationship between these variables in this specific subgroup of parents of children with ID who have a very complex profile of physical and psychological needs.

The effect of parental locus of control on subjective wellbeing shows that it is how parents perceive and make sense of their situation that is important, as families vary widely in what they consider challenging or not. The usefulness of screening caregivers of children with PMID for psychological distress was discussed in relation to early identification and support that may promote better outcomes. However, the evidence base for interventions that facilitate positive psychological outcomes for this group of parents is extremely limited and this urgently needs to be developed, perhaps starting with practice-based evidence.

A number of limitations of the present study were discussed in depth. Suggestions for further research that either overcomes these methodological limitations or builds on some of the interesting insights have been provided and this will hopefully result in further interest and research in this area for the benefit of these families.

References

- Aldridge, J. (2007). *Living with a seriously ill child*. London: Sheldon Press.
- Arvio, M., & Sillanpää, M. (2003). Prevalence, aetiology and comorbidity of severe and profound intellectual disability in Finland. *Journal of Intellectual Disability Research*, 47(2), 108-112.
- Baker, Blacher, & Olsson (2005). Preschool children with and without developmental delay: Behaviour problems, parents' optimism and well-being. *Journal of Intellectual Disability Research*, 49(8), 575-590.
- Beckman, P. J. (1991). Comparison of mothers' and fathers' perceptions of the effect of young children with and without disabilities. *American Journal on Mental Retardation*, 95, 585-595.
- British Psychological Society (2000). *Learning disability: Definitions and contexts*.
- Bunning, K. (2009). *Making sense of communication*. In J. Pawlyn & S. Carnaby, S. (Eds) *Profound intellectual and multiple disabilities: Nursing complex needs*. Chichester: John Wiley and Sons.

- Bytheway, B., & Johnson, J. (2008). *The social construction of carers*. In J. Johnson & C. De Souza (eds.) *Understanding health and social care*. London: Sage.
- Campis L. K., Lyman R. D., & Prentice-Dunn, S. (1986). The Parental Locus of Control Scale: Development and validation. *Journal of Clinical Child Psychology*, 15, 260-267.
- Chaney, R. H. (1996). Psychological stress in people with profound mental retardation. *Journal of Intellectual Disability Research*, 40(4), 305-310.
- Chavira, V., Lopez, S. R., Blacher, J., & Shapiro, J. (2000). Latina mothers' attributions, emotions, and reactions to the problem behaviors of their children with developmental disabilities. *Journal of Child Psychology and Psychiatry*, 41, 245-252.
- Cronbach, L.J. (1951). Coefficient alpha and the internal structure of tests. *Psychometrika*, 16(3), 297-334.
- Deater-Deckard, K. (2004). *Parenting stress*. London: Yale University Press.
- Department of Health (2001). *Valuing people: A new strategy for learning disability for the 21st Century*. London: Author.
- Douglas, H. (2007). *Containment and reciprocity: Integrating psychoanalytic theory and child development research for work with children*. London: Routledge.
- Edwards, M., & Titman, P. (2010). *Promoting psychological wellbeing in children with acute and chronic illness*. London: Jessica Kingsley.
- Emerson, E., Hatton, C., Felce, D. & Murphy, G. (2001). *Learning disabilities: The fundamental facts*. London: Foundation for People with Learning Disabilities.
- Emerson, E. & Hatton, C. (2009). *Estimating future need for adult social care services for people with learning disabilities in England: Centre for Disability Research report*. Retrieved 10 June 2011 from learningdisabilitycoalition.org.uk/download/CeDR_November.pdf
- Espie, C. A., Paul, A., McFie, J., Amos, P., Hamilton, D., McColl, J. H., Tarassenko, L., & Pardey, J. (1998). Sleep studies of adults with severe or profound mental retardation and epilepsy. *American Journal on Mental Retardation*, 103(1), 47-59.
- Fitton, P. (1994). *Listen to me: Communicating the needs of people with profound and multiple disabilities*. London: Jessica Kingsley.
- Foster, R. H., Kozachek, S., Stern, M., & Elsea, S. H. (2010). Caring for the caregivers: an investigation of factors related to well-being among parents caring for a child with Smith-Magenis syndrome. *Journal of Genetic Counselling*, 19, 187-198.
- Fotiadou, M., Barlow, J. H., Powell, L. A., & Langton, H. (2008). Optimism and psychological well-being among parents of children with cancer: An exploratory study. *Psycho-Oncology*, 17, 401-409.
- Glidden, L. M., Billings, F. J., & Jobe, B. M. (2006). Personality, coping style and well-being of parents rearing children with developmental disabilities. *Journal of Intellectual Disability Research*, 50(12), 949-962.

- Grove, N. (2007). Exploring the absence of high points in story reminiscence with carers of people with profound disabilities. *Journal of Policy and Practice in Intellectual Disabilities*, 4(4), 252-260.
- Hatton, C. (2002). Psychosocial interventions for adults with intellectual disabilities and mental health problems: A review. *Journal of Mental Health*, 11(4), 357-373.
- Hatzmann, J., Maurice-Stam, H., Heymans, H. S. A., & Grootenhuys, M. A. (2009). A predictive model of health related quality of life of parents of chronically ill children: The importance of care dependency of their child and their support system. *Health and Quality of Life Outcomes*, 7(72). Retrieved 10 October 2010 from hqlo.com/content/7/1/72
- Hogg, J. (1999). *People with profound intellectual and multiple disabilities: Understanding and realising their needs and those of their carers. Paper prepared for the Scottish Executive review of services for people with learning disabilities*. Retrieved 10 October 2010 from scotland.gov.uk/resource/doc/1095/0001692.pdf
- Hogg, J., & Lambe, L. (1988). *Sons and daughters with profound retardation and multiple handicaps attending schools and social education centres: Final report*. London: Mencap.
- Kearney, P. M., & Griffin, T. (2001). Between joy and sorrow: Being a parent of a child with a developmental disability. *Journal of Advanced Nursing*, 34(5), 582-592.
- Ketelaar, M., Volman, M. J. M., Gorter, J. W., & Vermeer, A. (2008). Stress in parents of children with cerebral palsy: What sources of stress are we talking about? *Child: Care, Health and Development*, 34(6), 825-829.
- Kirk, S. (1998). Families' experiences of caring at home for a technology-dependent child: a review of the literature. *Child: Care, Health and Development*, 24(2), 101-114.
- Kuster, P. A., & Badr, L. K. (2006). Mental health of mothers caring for ventilator-assisted children at home. *Issues in Mental Health Nursing*, 27, 817-835.
- Lambe, L. (1998). *Supporting families pp. 167-183*. In P. Lacey & C. Ouvry (eds.) *People with Profound and Multiple Learning Disabilities: A collaborative approach*. London: David Fulton.
- Lindsay, G., Davies, H., Band, S., Cullen, M. A., Cullen, S., Strand, S., Hasluck, C., Evans, R., & Stewart-Brown, S. (2008). *Parenting early intervention pathfinder evaluation*. Department for Children, Schools and Families.
- Lloyd, T. & Hastings, R.P. (2008). Psychological variables as correlates of adjustment in mothers of children with intellectual disabilities: Cross-sectional and longitudinal relationships. *Journal of Intellectual Disability Research*, 52, 37-48.
- Lloyd, T., & Hastings, R. P. (2009a). Hope as a psychological resilience factor in mothers and fathers of children with intellectual disabilities. *Journal of Intellectual Disability Research*, 53(12), 957-968.

- Lloyd, T., & Hastings, R. P. (2009b). Parental locus of control and psychological well-being in mothers of children with intellectual disability. *Journal of Intellectual and Developmental Disability*, 34(2), 104-115.
- Mencap (n.d.). *Facts about learning disability*. Retrieved 10 June 2011 from mencap.org.uk/all-about-learning-disability/about-learning-disability/facts-about-learning-disability
- Mencap & PMLD Network (n.d.). *About profound and multiple learning disabilities*. Retrieved 10 June 2011 from pmldnetwork.org/PMLD%20Definition%20factsheet%20-%20standard.pdf
- Michael, J. (2008) *Healthcare for all: Report of the independent enquiry into access to healthcare for people with learning disabilities*. Retrieved February 27, 2009, from: iahpld.org.uk/Healthcare_final.pdf
- Moss, S., Bouras, N., & Holt, G. (2000). Mental health services for people with intellectual disability: A conceptual framework. *Journal of Intellectual Disability Research*, 44(2), 97-107.
- Murphy, G. (1994). *Understanding challenging behaviour* (pp. 37-68). In E. Emerson, P. McGill & J. Mansell (eds.) *Severe Learning Disabilities and Challenging Behaviours: Designing high quality services*. London: Chapman and Hall.
- Nakken, H., & Vlaskamp, C. (2007). A need for a taxonomy for profound intellectual and multiple disabilities. *Journal of Policy and Practice in Intellectual Disabilities*, 42(2), 83-87.
- Neece, C. L., Green, S. A., & Baker, B. L. (2012). Parenting stress and child behavior problems: A transactional relationship across time. *American Journal on Intellectual and Developmental Disabilities*, 117(1), 48-66.
- Olsson, M. B., & Hwang, C. P. (2008). Socioeconomic and psychological variables as risk and protective factors for parental well-being in families of children with intellectual disabilities. *Journal of Intellectual Disability Research*, 52(12), 1102-1113.
- Ong, L., Chandran, V., & Peng, R. (2005). Stress experienced by mothers of Malaysian children with mental retardation. *Journal of Intellectual Disability Research*, 49(9), 657-671.
- O'Neil, E., Palisano, R. J., & Westcott, S. L. (2001). Relationship of therapists' attitudes, children's motor ability, and parenting stress to mothers' perceptions of therapists' behaviours during early intervention. *Physical Therapy*, 81, 1412-1424.
- Pawlyn, J., & Carnaby, S. (2009). *Profound intellectual and multiple disabilities: Nursing complex needs*. Chichester: John Wiley and Sons.
- Petry, K., Maes, B., & Vlaskamp, C. (2007). Support characteristics associated with the quality of life of people with profound multiple intellectual and multiple disabilities: The perspective of parents and direct support staff. *Journal of Policy and Practice in Intellectual Disabilities*, 4(2), 104-110.

- Pit-ten Cate, I. (2003). *Family adjustment to disability and chronic illness in children*. University of Southampton: Unpublished PhD thesis.
- Raina, P., O'Donnell, M., Schwellnuss, H., Rosenbaum, P., King, G., Brehaut, J., Russell, D., Swinton, M., King, S., Wong, M., Walter, S. D., & Wood, E. (2004). Caregiving process and caregiver burden: Conceptual models to guide research and practice. *BMC Pediatrics*, 4(1). Retrieved 10 June 2011 from biomedcentral.com/1471-2431/4/1
- Rousey, A. M., & Eyman, R. K. (1995). Probabilities of the development of ambulation for mobile children with moderate to profound mental retardation. *Mental Retardation*, 33, 322-327.
- Ruini, C., Ottolini, F., Rafanelli, C., Tossani, E., Ryff, C. D., & Fava, G. A. (2003). The relationship of psychological well-being to distress and personality. *Psychotherapy and Psychosomatics*, 72(5), 268-275.
- Seligman, M. E. P., & Csikszentmihalyi, M. (2000). Positive psychology: An introduction. *American Psychologist*, 55, 5-14.
- Sheehy, K., & Nind, M. (2005). Emotional well-being for all: Mental health and people with profound and multiple learning disabilities. *British Journal of Learning Disabilities*, 33, 34-38.
- Siman-Tov, A., & Kaniel, S. (2011). Stress and personal resource as predictors of the adjustment of parents to autistic children: A multivariate model. *Journal of Autism and Developmental Disorders*, 41, 879-890.
- Singer, G. H. S. (2006). Meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities. *American Journal on Mental Retardation*, 111(3), 155-169.
- Stewart-Brown, S., Tennant, A., Tennant, R., Platt, S., Parkinson, J., & Weich, S. (2009). Internal construct validity of the Warwick-Edinburgh Mental Well-being Scale (WEMWBS): A Rasch analysis using data from the Scottish Health Education Population Survey. *Health and Quality of Life Outcomes*, 7(15). Retrieved 10 June 2011 from hqlo.com/content/7/1/15
- Tennant, R., Fishwick, R., Platt, S., Joseph, S., & Stewart-Brown, S. (2007). *Monitoring positive mental health in Scotland: Validating the Affectometer 2 scale and developing the Warwick Edinburgh Mental Well-being Scale for the UK*. Edinburgh: NHS Health Scotland.
- Wallander, J.L., & Varni, J.W. (1998). Effects of pediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry*, 39(1), 29-46.

CHAPTER 4: EXTENDED METHOD

Following on from the journal article summarising the research study, this section describes the method of the study in further detail. This includes information regarding the study design, procedure, measures employed and sample demographics. Ethical issues are also considered, in addition to data protection and storage considerations. Finally, planned analyses of the data are outlined.

Ethical considerations

The study received ethical approval from the University of Edinburgh School of Health in Social Science Ethics Committee; level one scrutiny was deemed appropriate. In response to the ethical issue regarding the potentially distressing nature of the study, this was highlighted in the information provided to potential participants prior to them deciding whether to give their informed consent to take part or not (see Appendix 6). In addition to this, information was provided about how participants could seek further support. Contact details for the Research Director for the School of Health in Social Science at the University of Edinburgh were provided in the event of an individual wanting to speak to someone independent from the study.

Design

The study employed a cross-sectional questionnaire survey design. Repeated measures analyses were used to explore the relationships among the three variables under investigation², using data from a single sample. The study involved completion of the questionnaire items either in the format of a hard copy paper booklet or online via a secure website.

Data protection and storage

Anonymity was ensured by asking participants only for non-identifiable information. In the participant information provided at the beginning of the survey, parents and carers were made aware that they did not have to provide information that they felt would result in themselves or their family being identifiable (e.g. rare syndromes). A participant identifier (PID) was written onto paper copies of the questionnaire booklets (e.g. P01, P02) so that individual responses could be linked together. For those completing the survey online, the last section of the online survey asked participants to generate a unique PID by combining the first two letters of their surname, the last two letters of their postcode and their month of birth (e.g. 'smab04').

² Based on power calculations, a pragmatic decision was taken to limit the number of variables investigated to two predictor variables and one criterion variable. This decision was based on the potential difficulty identified in accessing the study population and expectations of recruitment difficulties due to the nature of participants' caring demands.

Raw data (in the form of participants' coded responses) were held on the Bristol Online Survey secure server, a UK-based service that the University of Edinburgh subscribes to for the purpose of hosting surveys in line with the Data Protection Act 1998. These were then transferred directly to a password protected Microsoft Excel database, in addition to an SPSS database on the author's password protected home computer. Completed paper questionnaires that had been returned were stored in a locked filing cabinet at the author's place of employment and paper questionnaire booklets were destroyed once data had been entered into the results database.

Participants

Participants comprised 101 parents and family carers who were self-selected and were recruited to the study via two different routes. Two local independent schools with special provision for children and young people with PMID run by Capability Scotland and Royal Blind were approached initially and they agreed to distribute questionnaires to parents and carers. Alongside this, support groups, voluntary organisations and charities for children with disabilities, physical health conditions (including genetic conditions, chromosomal abnormalities and syndromes) and/or complex needs were also approached and asked to share information about the study, including the online questionnaire survey web address. A full list of organisations that supported the study can be found in Appendix 7, in addition to a list of forums, email lists and social media sites that were used to recruit participants.

Inclusion and exclusion criteria

Inclusion criteria were: being a parent or carer and having at least one child with PMID. Exclusion criteria were: being a carer paid in an employment role (foster carers were still included) and the child with PMID being over 19. However, parents of children with PMID up to the age of 23 participated in the study. This was deemed close enough to the cutoff age for their data to be included in the study as being relevant.

Group definitions

Different terms are often used to describe PMID, so the participant information section clarified that this study was adopting the criteria published in Mencap and the PMLD Network's leaflet on the definition of PMID (n.d.). It highlighted that we were referring to children and young people under 19 who needed high levels of support with most aspects of daily life due to having more than one disability, including:

- (i) *a profound intellectual disability*
- (ii) *significant communication difficulties*
- (iii) *sensory impairment (e.g. hearing, vision)*
- (iv) *physical disabilities and mobility problems*
- (v) *complex health needs (e.g. enteral/parenteral feeding, ventilation/CPAP, etc)*
- (vi) *mental health difficulties*

Given the self-selecting nature of participation, those who took part were people who viewed themselves as a parent or carer. The term 'carer' was purposefully not defined, given the multitude of different conceptualisations of this role (Bytheway & Johnson, 2008) and in order to capture data from a broad range of individuals in a caring role.

Demographic characteristics of the caregivers who participated are summarised in Table 5. Caregivers in this study were predominantly white (97%), female (96%), a parent of the child (96%), and married, in a civil partnership or cohabiting (71%). Most caregivers were aged 41-50 years (40%), followed by the 31-40 age group (32%). In terms of socio-economic status, the majority were employed or in education (52%), educated to undergraduate level (30%), and owner-occupiers of their homes (67%). Almost half of the caregivers surveyed (45%) reported that they had their own health issues (e.g. back pain, depression, high blood pressure and psoriasis). An overview of these is provided in Table 6.

Demographic characteristics of the children with PMID are summarised in Table 7. Twenty per cent of children with PMID were living in a one-parent household. At least sixty per cent of the children with PMID had siblings living in the same family home, although thirteen per cent of the sample did not provide information for this item. Thirty-seven per cent of the overall sample had one sibling at home, nineteen per cent had two siblings at home and eight per cent had three or more siblings at home. Children with PMID had a range of conditions, including genetic and chromosomal abnormalities (e.g. Cri du Chat syndrome, Phelan McDermid syndrome and Di George syndrome), cerebral palsy, epilepsy, scoliosis, gastrointestinal issues and hearing or vision impairment. An overview of these is provided in Table 8.

Table 5: Demographic characteristics of caregivers

		Percentage of sample N= 101
Age in years	≤ 20	1
	21-30	8
	31-40	32
	41-50	40
	51-60	18
	61-70	1
Gender	Male	4
	Female	96
Caregiving role	Parent	96
	Foster carer	2
	Other family carer	2
	Other	1
Ethnicity	White British	45
	White Scottish	33
	White Irish	7
	White Other	13
	Black African	1
	Other Ethnicity	2
Employment status	Not employed / retired	49
	Employed part-time	32
	Employed full-time	17
	Full-time student	2
	Part-time student	1
Highest level of education	Undergraduate	30
	GCSE/Standard Grade/NVQ/SVQ	25
	Postgraduate	19
	AS/Level/Higher/A Level	16
	No formal qualifications	6
	Other	5
Living situation	Owner occupier	67
	Council rented	16
	Private rented	9
	Other rented	5
	Other	4
Marital status	Married/civil partnership	70
	Single	10
	Cohabiting	9
	Divorced	8
	Widowed	2
	Other	29
Health issues n= 89	Yes	45
	No	43

Table 6: Health conditions of caregivers

Anxiety	Irritable bowel syndrome
Back pain	Lupus
Breast cancer	Osteoarthritis
Depression	Previous stroke
Fibromyalgia	Psoriasis
High blood pressure	

Table 7: Demographic characteristics of children with PMID

		n= 88
Age in years	Mean	10.36
	SD	5.85
	Range	1-23
		Percentage of sample N= 101
Gender	Male	59
	Female	40
Ethnicity	White British	45
	White Scottish	31
	White Other	10
	White Irish	8
	Mixed	2
	Other	2
	Asian Indian	1
	Black African	1

Table 8: Medical conditions and presenting problems of children with PMID

Agenesis of corpus callosum	Hypoparathyroidism
Attention problems	Hypotonia
Autistic features	Incontinence
Cardiac problems	Other chromosome disorders (e.g. deletions, trisomies, tetrasomies, translocations and disorders of chromosomes)
Cerebral palsy	Periventricular heterotopia / leukomalasia
Cri du Chat syndrome	Phelan McDermid syndrome
Di George syndrome	Pituitary gland dysfunction
Eczema	Quadriplegia
Epilepsy	Respiratory problems
Gastrointestinal problems (e.g. reflux)	Scoliosis
Hearing impairment	Speech/communication problems
Hydrocephalus	Visual impairment
Hypohidrotic ectodermal dysplasias	

Procedure

Information was provided to potential participants regarding the nature and purpose of the study so that parents and carers could decide whether they wanted to participate or not (see Appendix 6). Participation was entirely voluntary and the participant information section highlighted that whether or not they took part would not affect the services families received in any way. Completion and submission of the questionnaires were deemed as giving informed consent for responses to be used for the purpose of the study and participants were made aware of this.

The survey was made available both as a hard copy paper booklet and also as an online survey accessed via a secure website. Participants were first asked to provide demographic information, the Parental Locus of Control Scale - Revised (PLOC-R; Lloyd & Hastings, 2009) then followed, in addition to the Positive Gain Scale (PGS; Pit-ten Cate, 2003) and finally the Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS; Stewart-Brown *et al.*, 2009; Tennant *et al.*, 2007). Space was provided for any comments or feedback parents wished to share in relation to their experiences or the research study itself.

Participants were reminded at the end of the questionnaires of their right to withdraw their data from the study at any time. They were also thanked for their valued time and provided with contact details for the author, academic supervisor and a person linked to the university but independent from the study in the event they wished to get further information about the study or sources of support.

Measures

In addition to demographic information, three self-report questionnaires were used to collect quantitative data relating to the three variables under investigation. Copies of each measure are included in Appendices 8-10. The measures (described in more detail below) were selected primarily on the basis of having robust psychometric properties and having been used in previous research with parents of children with ID and/or chronic health issues (*cf.* Lloyd & Hastings, 2009; Pit-ten Cate, 2003). Despite there being a range of self-report measures available for assessing the cognitive domains of interest, there were very few that had been used already with this population, thus limiting the selection deemed appropriate.

Parental Locus of Control Scale - Revised

The Parental Locus of Control Scale (see Appendix 8) - revised version (Lloyd & Hastings, 2009) was used to collect data relating to parents' locus of control within the context of the parent-child relationship. The scale has been adapted specifically for parents of children with ID (*cf.* Campis, Lyman, & Prentice-Dunn, 1986 for original version). The revised scale includes 42 statements that parents respond to in terms of how much they

agree or not, using a four-point Likert-type scale. Subscales include items that explore parental efficacy, responsibility, belief in fate/chance and child control over parents' life.

Lloyd and Hastings (2009) used an item-reduction procedure to develop a robust revised version of the measure with acceptable alpha levels (parental efficacy .69, 8 items; parental responsibility .81, 10 items; child control .70, 5 items; fate/chance .67, 9 items; parent control .82, 10 items). The Cronbach's alpha for the total PLOC-R score (general internal-external orientation) based on these 42 items was also good at .81.

For the purpose of the current study, the PLOC-R was adapted slightly. A five-point response scale was used instead of the original four-point scale, given that the other two questionnaires in the study included five-point response scales. An additional mid-point option of 'not sure' was added to the existing 'strongly agree/agree/disagree/strongly disagree' response options. Using the five-point scale, the internal consistency of the PLOC-R was good, with a Cronbach's alpha of .92.

For the current study, a PLOC-R total score was used as a predictor variable to investigate the relationship between parental locus of control and parental subjective wellbeing. The maximum score on this scale is 210, with a high score indicating an external locus of control in relation to the respondent's parenting of their child.

Positive Gain Scale (PGS)

The Positive Gain Scale (PGS; Pit-ten Cate, 2003; see Appendix 9) was used to assess positive experiences associated with raising a child with a disability. The measure consists of seven items; five relating to the perceived benefits for the parent of raising a child with a disability and two focusing on what the family has gained. Responses to each of the seven statements are given using five-item Likert type scale, ranging from 'strongly agree' to 'strongly disagree'. Preliminary research findings have indicated that the PGS has face and content validity and a Cronbach's alpha coefficient of .79 for parents of children with hydrocephalus and spina bifida (Pit-ten Cate, 2003). A Cronbach's alpha co-efficient for the total positive gain score of 0.80 has been obtained in a study where the PGS was used with parents of children with ID (MacDonald *et al.*, 2010). For the current study, the PGS demonstrated an acceptable level of internal consistency, with a Cronbach's alpha of .70.

A PGS total score was used as a predictor variable in the current study to investigate the relationship between parents' positive perceptions of their child with PMID and parental subjective wellbeing. The maximum score on this scale is 35, with a low score indicating higher positive gains reported by parents.

Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS)

The Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS; Stewart-Brown *et al.*, 2009; Tennant *et al.*, 2007; see Appendix 10) was used to collect data relating to parents' subjective psychological wellbeing. It is a brief, 14 item scale covering most aspects of positive mental health (positive thoughts and feelings) and psychological wellbeing currently in the literature, including both hedonic and eudaimonic perspectives. All items are worded positively and address aspects of positive mental health. Responses to each of the 14 statements are given using five-item Likert type scale, ranging from 'none of the time' to 'all of the time'.

The scale has been validated for use in the UK with those aged 16 and above and was standardised with both student and general population samples, in addition to focus group exploration of acceptability (Stewart-Brown *et al.*, 2009; Tennant *et al.*, 2007). These two studies described the psychometric properties of the WEMWBS in detail. Scores derived from the student and population samples showed a single underlying factor, interpreted to be mental wellbeing, with low levels of social desirability bias and expected moderate correlations with other scales of wellbeing (Stewart-Brown *et al.*, 2009; Tennant *et al.*, 2007). The WEMWBS has been found to be psychometrically robust in terms of principal components factor analysis, construct validity, internal consistency, test-retest reliability, response bias face/content validity and Rasch analysis (NHS Health Scotland, 2008). For the current study, internal consistency was high, with a Cronbach's alpha of .87.

As there is no specific measure of subjective wellbeing specifically designed for parents of children with ID, the WEMWBS was selected due to its general nature. The measure has been used to assess parental wellbeing in an evaluation of parenting interventions (Lindsay *et al.*, 2008). It was also selected on the basis of the measure being positively focused on wellbeing as opposed to problem-oriented.

A WEMWBS total score was used to explore relationships between positive child perceptions, locus of control and parental subjective wellbeing. The maximum score on this scale is 70, with higher scores representing higher subjective psychological wellbeing.

Treatment of multiple responses and missing data

Some of the parents who completed the hard copy questionnaire booklets gave more than one response, wrote 'N/A' as their response or left questions blank. This was not an issue for the online version of the survey, as it was a requirement in order to progress to the next page and submit responses that all items had been completed with one response. Where multiple responses were given, the mid-point value was used as the response. Items which had been left blank or answered as 'N/A' were treated as missing data. These were given discrete values ('999') in the SPSS database in order to label them and were excluded

listwise for all analyses (n=12) in order to minimise the impact of the missing data on the results, given the within-participant design of the study.

Data analysis

Raw data collated from hard copy paper questionnaire booklets were entered directly into an SPSS database, using version 19.0 for MAC OS X. Raw data from the online survey were extracted in .csv format, saved in a Microsoft Excel document and an Excel formula was employed to reverse score any items that required this before being transferred directly to the same SPSS database as the booklet derived data. Scores for all participants were included in the analyses (N= 101), except where responses were missing.

Planned analyses

Correlational analyses were planned in order to explore any relationships between the variables. Based on the findings of the systematic review, multiple regression analysis was also selected to investigate predictive power of parental locus of control and realisation of the positive gains of having a child with PMID, in relation to parental subjective wellbeing.

A priori power calculations using G*Power software (Faul *et al.*, 2007; 2009) for a multiple regression analysis with an alpha level of .05, two predictor variables, an anticipated effect size of 0.15 (medium) and a statistical power level of 0.8 indicated a minimum required sample size of 67 participants.

CHAPTER 5: EXTENDED RESULTS

This chapter describes initial exploration of the data and the inferential statistical analyses undertaken to examine whether the data supported the research hypotheses outlined in Chapter Three. All analyses are summarised in Tables 9-11. Figures 2-4 reveal the distribution of the raw data for the predictor and criterion variables. Figure 5 shows the distribution of the transformed data for PGS total scores, which is described in further detail later in the chapter.

Exploration of data

In order to determine whether the planned inferential analyses would be appropriate, it was important to explore the data to gain an overview of their characteristics and to ascertain whether parametric assumptions had been met for the variables under investigation. Table 9 summarises measures of central tendency, dispersion and the shape of the distribution that were calculated as part of the initial stage of data exploration.

Table 9: Key variable measures of central tendency, dispersion, skewness and kurtosis

	Parental locus of control PLOC-R total score	Positive gains PGS total score	Parental subjective wellbeing WEMWBS total score
Mean	113.42	23.51	39.49
Mode	113.50	26.00	36.00
Median	115.00	26.00	39.00
Range	66-153	1-35	22-60
<i>SD</i>	16.47	8.56	7.57
Skewness z-score ^a	-0.46	4.36	0.80
Kurtosis z-score ^a	0.34	1.39	0.34

^a Calculated using formula from Field (2009)

In order to determine whether parametric inferential analyses were appropriate or not, histograms for the predictor and criterion variables were consulted (see Figures 2-4) to gain an overview of the shape of the distributions. Skewness and kurtosis z-scores were also examined, and a Shapiro-Wilks test was used to explore whether the data differed significantly from a normal distribution for each variable.

Parental subjective psychological wellbeing

Figure 2 shows the frequency distribution of total scores on the WEMWBS and suggests a normal distribution, which was also indicated by the skewness and kurtosis z-score values for data relating to this variable (see Table 9). The Shapiro-Wilks statistic for WEMWBS data was non-significant, therefore the assumption of normality was fulfilled for the criterion variable.

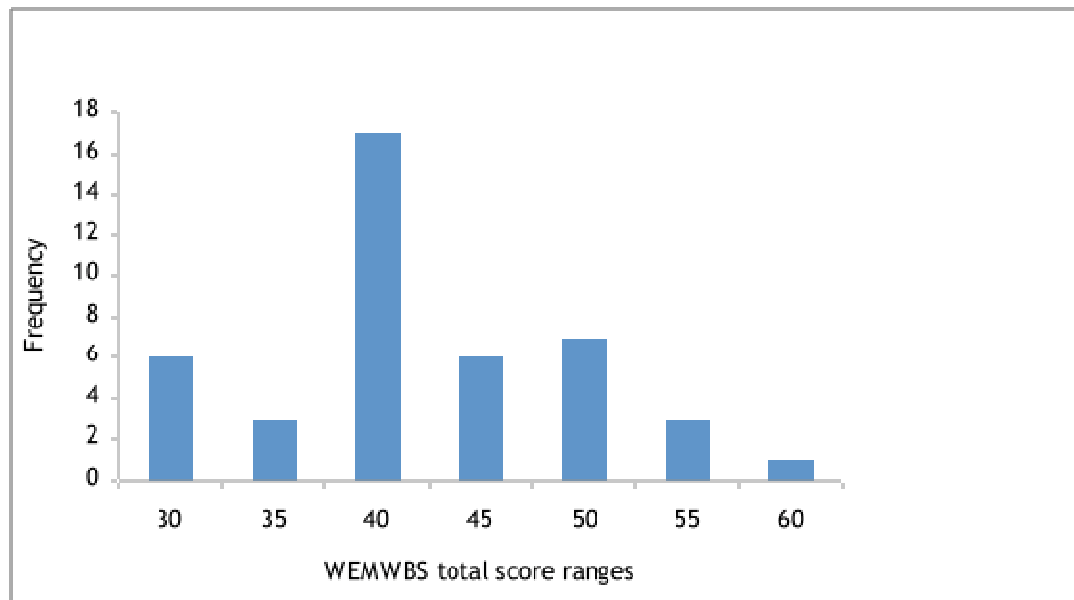


Figure 2: Histogram showing frequency distribution of WEMWBS total scores

Parental locus of control

Figure 3 shows the frequency distribution of total scores on the PLOC-R and suggests a normal distribution, which was again also indicated by the skewness and kurtosis z-score values relating to this variable (see Table 9). The Shapiro-Wilks statistic for PLOC-R data was non-significant, therefore the assumption of normality was fulfilled for this predictor variable.

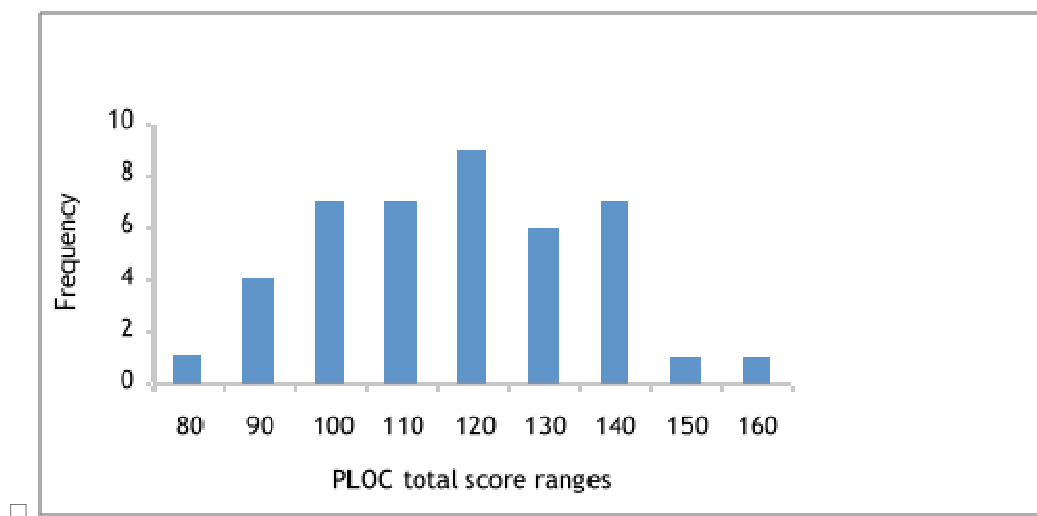


Figure 3: Histogram showing frequency distribution of PLOC-R total scores

Positive gains of having a child with a disability

Figure 4 shows the frequency distribution of total scores on the PGS, revealing a pile-up of scores on the left-hand side of the distribution. This means that the majority of parents' responses indicated that they were able to identify positive gains of having a child with PMID. Possible explanations for this particular pattern of results will be explored in more detail in the Discussion chapter.

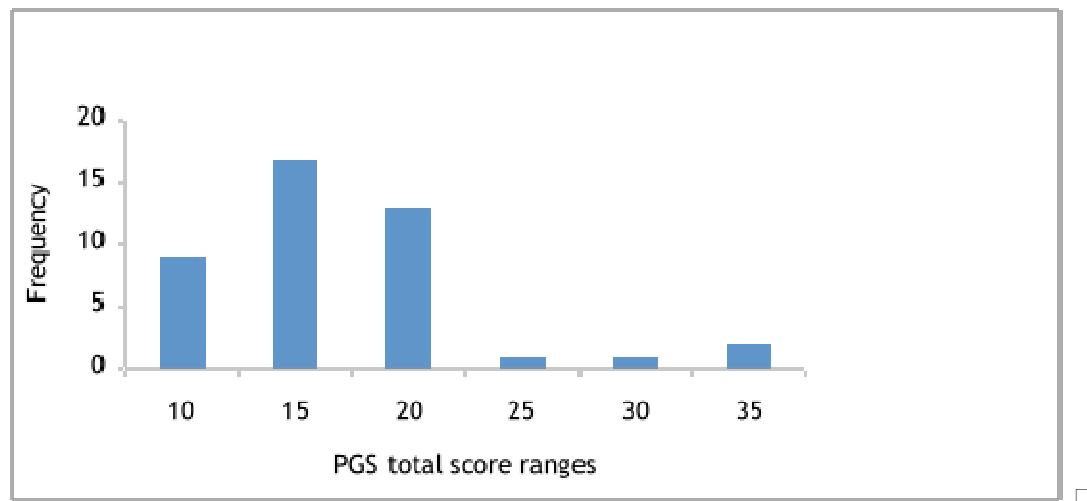


Figure 4: Histogram showing frequency distribution of PGS total scores

Although the kurtosis z-score value for this variable (see Table 9) is within the normal range (Field, 2009), the skewness z-score value clearly indicates that the distribution is not normal. The Shapiro-Wilks statistic was significant for data relating to this variable ($W = .909$, $df = 102$, $p < .001$), therefore, the data for this variable did not fulfill assumptions for normal distribution. A natural log transformation process was used in addition to applying a constant to the data (-1) for this variable in order to yield a normal distribution so that the planned regression analyses would still be an option. Figure 4 shows the new distribution of the data for this variable, following data transformation.

Following transformation of the data for the PGS scores (see Figure 5), data for the two predictor variables and the criterion variable were deemed to fulfill assumptions for the planned multiple regression analysis. These included the data being interval level for all variables, a minimum number of 10 observations per predictor variable (Brace *et al.*, 2009), a normal distribution, a linear relationship between the criterion and predictor variables, no collinearity between the predictor variables, reliably demonstrated measurement without error (*i.e.* Cronbach's alphas of .7-.8 are acceptable) and homogeneity of variance (also referred to as homoscedasticity; Field, 2009).

□

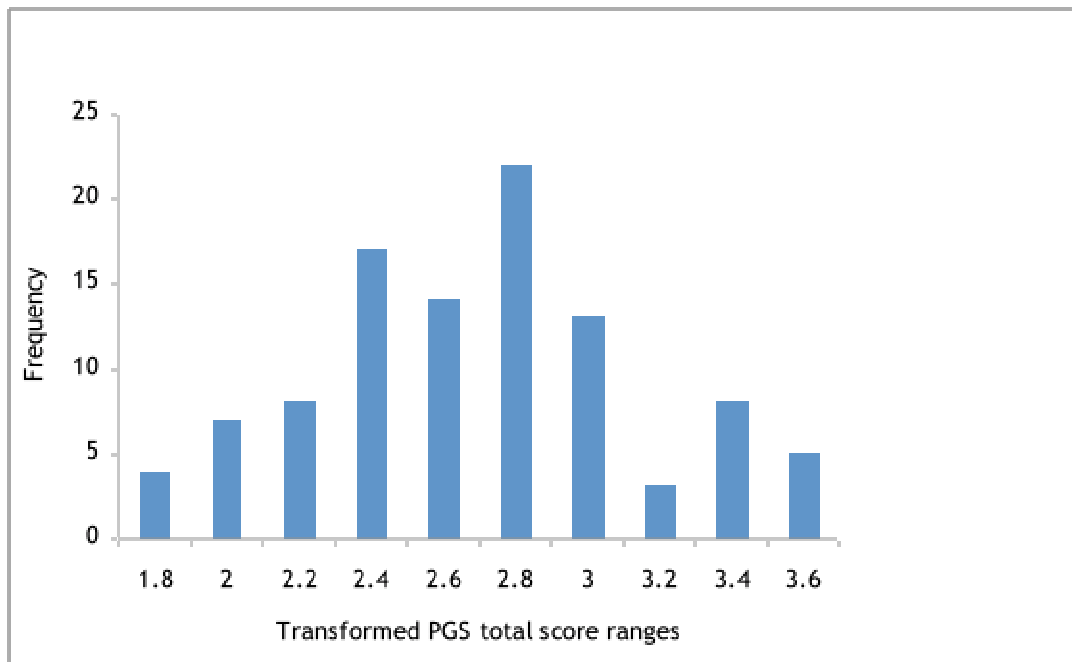


Figure 5: Histogram showing frequency distribution of Log-1 transformed PGS scores

Inferential analyses

Two sets of parametric inferential analyses were undertaken using IBM SPSS Statistics Version 19.0 for Mac OS X. These were undertaken using the original data from two of the key variables (PLOC-R total scores and WEMWBS total scores). Transformed data for the third key variable (PGS total scores) was used in all of the inferential analyses to enable parametric analyses to be conducted. First, relationships among the key variables (parental locus of control and realisation of positive gain from having a child with PMID and parental subjective wellbeing) were examined using Pearson bivariate correlational analyses. Second, a linear multiple regression analysis employing the enter method was used to explore the extent to which the two types of parental cognitions could predict parental subjective wellbeing.

Relationships among key variables

Table 10 summarises the parametric bivariate correlational analyses used to explore the relationships between scores on the PLOC-R, PGS and WEMWBS. The only statistically significant relationship to emerge among the key variables was a significant negative correlation between PLOC-R total scores and WEMWBS total score, $r = -.295$, $p = .003$. This means that higher scores on the PLOC-R indicating an external parental locus of control were related to lower parental subjective wellbeing scores, which related to lower psychological wellbeing. There were no statistically significant relationships observed between PLOC-R total scores and PGS transformed total scores, nor between PGS transformed total scores and WEMWBS total scores (see Table 10).

Table 10: Bivariate and point-biserial Pearson correlations between variables

	Parental health status	Child age	PLOC-R total score	PGS total score log -1 transformed	WEMWBS total score
Parental health status		$r = .213$ $p = .047$	$r = -.054$ $p = .590$	$r = .604$ $p < .001$	$r = -.150$ $p = .135$
Child age	$r = .213$ $p = .047$		$r = .067$ $p = .535$	$r = .009$ $p = .937$	$r = .031$ $p = .771$
PLOC-R total score	$r = -.054$ $p = .590$	$r = .067$ $p = .535$		$r = .129$ $p = .199$	$r = -.295$ $p = .003$
PGS total score log -1 transformed	$r = .604$ $p < .001$	$r = .009$ $p = .937$	$r = .129$ $p = .199$		$r = -.153$ $p = .126$
WEMWBS total score	$r = -.150$ $p = .135$	$r = .031$ $p = .771$	$r = -.295$ $p = .003$	$r = -.153$ $p = .126$	

Relationships with other variables

Parametric bivariate correlations were also used to explore the relationship between child age and the key variables in the study. However, no statistically significant relationships were observed between child age and PLOC-R total score, WEMWBS total score and PGS transformed total scores (see Table 10). Furthermore, Pearson point-biserial correlational analyses were undertaken to explore whether parental health status (categorised into two groups: whether they had identified health issues or not) was associated with any of the key variables, in addition to child age. As Table 10 shows, statistically significant associations were revealed between parental health status and both child age ($r = .213$, $p = .047$), and the PGS transformed total scores ($r = .604$, $p < .001$).

Cognitive factors as predictors of parental psychological wellbeing

A linear multiple regression analysis employing the enter (also referred to as simultaneous) method was used to determine whether each of the respective cognitive factors (parental locus of control and recognition of positive gains of having a child with PMID) could predict parental subjective wellbeing (as measured by the WEMWBS). Data relating to parents' total scores on the PLOC-R and transformed data relating to total scores on the PGS were entered as a block of predictor variables at Step 1. They were entered this way as the analyses were exploratory (Brace *et al.*, 2009).

Regression analysis (see Table 11) revealed that parental locus of control significantly predicted parental subjective wellbeing, $B = -.279$, $t(2,99) = 9.419$, $p = .005$. Parental locus of control explained around eight per cent of the variance in WEMWBS scores, adjusted $R^2 = .081$, $F(2,99) = 5.474$, $p = .006$. However, the regression analysis also revealed that realisation of positive gains of having a child with PMID did not significantly predict parental subjective wellbeing and did not account for any of the variance in WEMWBS scores.

Table 11: Multiple regression analysis to predict parental subjective wellbeing

Variables	<i>B</i>	<i>SE B</i>	<i>B</i>	<i>t</i>	<i>p</i>
Step 0					
Constant	59.521	6.319		9.419	<0.001
Step 1					
PLOC-R	-0.131	0.045	-0.279	-2.895	0.005
PGS-LT	-1.977	1.649	-0.115	-1.199	0.233

B = unstandardised beta coefficient, *SE B* = standard error,
B = standardised beta coefficient, *t* = test statistic, *p* = significance value
PLOC-R = Parental Locus of Control Scale - Revised, PGS-T = Positive Gain Scale - log transformed data

Summary of results

The mean WEMWBS total score of this sample was more than 10 points below the general population mean reported by Tennant *et al.* (2007) and was also less than that reported for a group of parents of children who did not have a disability taking part in an early intervention project (Lindsay *et al.*, 2008). The only statistically significant relationship to emerge among the three key variables under investigation was a significant inverse correlation between parental locus of control and parental subjective wellbeing (Pearson's $r = -.294$, $p = .003$). This means that parents with higher scores on the PLOC-R (indicating a more external parental locus of control) had lower scores on the WEMWBS (indicating lower parental subjective wellbeing). The regression analysis investigated this relationship further and demonstrated that parental locus of control was a statistically significant predictor of parental subjective wellbeing ($B = -.279$, $t(2,99) = 9.419$, $p = .005$), accounting for around eight per cent of the variance in WEMWBS scores, $R^2 = .081$, $F(2,99) = 5.474$, $p = .006$.

CHAPTER 6: EXTENDED DISCUSSION

This chapter summarises the findings from the previous results chapter and considers these in detail, with reference to the hypotheses outlined in Chapter Three, the wider literature and clinical implications. Limitations of the study are also explored, in addition to directions for future research.

Parental locus of control and subjective psychological wellbeing

It was hypothesised that self-reported parental locus of control would be predictive of self-reported parental subjective wellbeing in the sample of caregivers of children with PMID surveyed. A statistically significant inverse relationship between parental locus of control (measured using PLOC-R total scores) and parental subjective wellbeing (measured using WEMWBS total scores) emerged, with a medium effect size ($r = -.295$, $p = .003$).

Higher total scores on the PLOC-R indicated a more external parental locus of control and higher total scores on the WEMWBS corresponded with increased subjective wellbeing. Therefore, in this group of caregivers of children with PMID, a more external parental locus of control was related to lower levels of parental subjective wellbeing. This is in line with previous research findings highlighting a relationship between parental locus of control and negative parent psychological outcomes (e.g. higher levels of depression and isolation: Dunn *et al.*, 2001; parental stress: Hassall *et al.*, 2005; Lloyd & Hastings, 2009; depression and anxiety: Lloyd & Hastings, 2009; and maladaptive coping: Glenn *et al.*, 2009). The findings of the present study provide further evidentiary support for the relationship between parental locus of control and wellbeing in parents who have a child with ID. They also emphasise the importance of cognitive and other factors internal to the parent that mediate the relationship between the stressor of having a child with a disability and psychological outcomes such as coping and adjustment, as outlined in several key theoretical models of this process (Lazarus & Folkman, 1984; Lazarus, 1991; McCubbin & Patterson, 1983; Wallander, Varni, Babani, DeHaan, *et al.*, 1989). Furthermore, this is the first known study to investigate the relationship between these variables in this specific subgroup of parents of children with ID who have a very complex profile of physical and psychological needs.

Further exploration of the relationship between parental locus of control and subjective wellbeing using regression analysis revealed that, as hypothesised, parental locus of control significantly predicted the subjective wellbeing of parents of children with PMID ($\beta = -.279$, $t(2,99) = 9.419$, $p = .005$). Parental locus of control as a predictor accounted for around eight per cent of the variance in WEMWBS scores, $R^2 = .081$, $F(2,99) = 5.474$, $p = .006$. This concurs with previous findings reported in the systematic review that highlighted the importance of parental locus of control to a number of parent psychological outcomes (e.g. Dunn *et al.*, 2001; Hassall *et al.*, 2005; Lloyd & Hastings, 2009; Lloyd & Hastings, 2009; Glenn *et al.*,

2009). These findings also fit with the theoretical models outlined in earlier chapters that conceptualise the stress-coping process and aim to explain the individual variation in parental outcomes (Lazarus & Folkman, 1984; Lazarus, 1991; McCubbin & Patterson, 1983; Wallander *et al.*, 1989). The findings of the current study add to the evidence base, as they have demonstrated similar effects of parental cognitions to studies of parents of children with other types of disability, with a specific subgroup of parents. Furthermore, the study adopted a strengths based approach focusing on subjective wellbeing rather than parental stress, which has also never been investigated with this population of parents.

Recognition of positive gains of having a child with PMID and subjective psychological wellbeing

It was hypothesised that self-reported parental recognition of the positive gains of having a child with PMID would be predictive of self-reported subjective wellbeing in the sample of caregivers of children with PMID surveyed. However, the results of the current study did not support this hypothesis as no statistically significant relationship emerged between recognition of positive gains of having a child with PMID (measured using PGS total scores) and parent subjective wellbeing (measured using WEMWBS total scores). Further exploration using regression analysis did not reveal recognition of positive gains as a significant predictor and it did not account for a significant proportion of the variance in parental subjective wellbeing. This suggests that in this sample, being able to recognise positive aspects of having a child with PMID did not influence wellbeing, as has been observed in other studies including more heterogeneous groups of parents of children with ID (e.g. Lloyd & Hastings, 2009a; Rapanaro *et al.*, 2008).

There are several possible explanations that may account for the pattern of these results. First, the current study used an unpublished quantitative measure of the positive gains associated with having a child with a disability (Pit-ten Cate, 2003). Although this measure asks parents to rate general statements relating to positive aspects of the experience of having a child with a disability (all PGS items can be found in Appendix 9), the measure was originally used with parents of children with hydrocephalus and spina bifida, both of which can, but do not necessarily, result in ID. Thus, the pattern of results with these populations of parents may differ from the findings for parents of children with PMID in this study due to the differences in the nature of the children's functioning. If a child has a physical but not an intellectual disability, this is likely to make the parenting experience differ from having a child with multiple physical and profound intellectual disabilities.

The distribution of the raw total scores of parents in this group (see Figure 4) revealed a pile-up of scores on the left hand side of the distribution that differed significantly from a normal distribution ($W = .909$, $df = 102$, $p < .001$). The trend for this group of caregivers to achieve lower total scores on this measure was clearly evident, with lower scores on the

PGS representing higher identification of positive gains of having a child with a disability. Caregiver total scores on the PGS almost covered the full range that can be achieved on the seven item measure (1-35), indicating that extreme floor effects were not demonstrated. However, as a group, these caregivers achieved a relatively high mean total score of 23.51 (SD= 8.56) and the median value was similar at 26. Higher scores on this measure indicate recognition of fewer positive gains, thus there were some parents in this sample that found it difficult to see positive aspects of having a child with PMID. Higher scores on this measure indicate recognition of fewer positive gains, thus there were some parents in this sample that found it difficult to see positive aspects of having a child with PMID.

One possible explanation for the pile up of scores on the left of the distribution, indicating more parental recognition of positive gains, is that caregivers were exhibiting a tendency to endorse statements that would be socially valued (e.g. ‘Since having this child, my family has become closer to one another’), also known as the social desirability effect. Robinson and Anderson (1983) caution against reliance on only self-report measures in studies of parenting and family functioning for this reason. Indeed, given the fact that caregivers of children with PMID seldom seem to be asked how they feel about the experience of having a child with such complex needs (Fitton, 1994) and the paucity of research investigating their experiences, it is plausible that they may feel uncomfortable expressing thoughts that may not be socially desirable.

The way positive gains were measured may also account for the pattern of findings observed. Previous studies that have investigated positive gains or perceived benefits of having a child with a disability have reported using quantitative scales (e.g. Positive Contributions Scale; Lloyd & Hastings, 2009a) or qualitative questionnaires (e.g. Rapanaro *et al.*, 2008) that differ to the PGS employed in the current study. Although the PGS has been used in a similar study (MacDonald *et al.*, 2010), it was used in a slightly different way. Both positive gains and negative consequences of raising a child with ID were investigated by MacDonald *et al.* (2010) and these were used as dependent variables, not as predictor variables as was the case in the current study. These methodological differences may go some way to explaining the different pattern of findings in this study. It must also be noted that the PGS only has seven items, whereas the other measures employed in the current study consisted of at least twice as many items. It may be that this measure does not include enough items to be sensitive enough to differences between individuals within a group of parents who likely all value and see the positives in having such special children.

Influence of child age and parental health status

Although recognition of positive gains was not associated with parental subjective wellbeing or parental locus of control, a statistically significant relationship was revealed between transformed PGS total scores and whether the parent had identified that they had

a health issue or not ($r = .604, p < .001$). This meant that parents who identified themselves as having a health issue were more likely to achieve higher scores on the PGS, indicating recognition of fewer positive gains. Parents with health issues may find it more difficult to see the positive gains of having a child with PMID because it is likely to impact negatively on their physical and mental health, although we cannot infer the direction of causality.

Furthermore, a statistically significant association between parental health status and child age was also revealed ($r = .213, p = .047$), indicating that parents with older children tended to have significantly more identified health issues, perhaps compounding this issue. This would make sense in terms of the cumulative burden of the physical and emotional care demands associated with parenting a child with PMID needs (Nakken & Vlaskamp, 2007). However, child age was not significantly associated with any of the other variables investigated.

Influence of variables that were not investigated

Each of the four key theoretical models outlined in earlier chapters highlighted the multi-dimensional and transactional nature of the process of responding to stress (Lazarus & Folkman, 1984; Lazarus, 1991; McCubbin & Patterson, 1983; Wallander, Varni, Babani, DeHaan, *et al.*, 1989). Key variables identified in these stress-coping models that influence parental and family outcomes include cognitive, emotional and behavioural responses to the stressor. These can be at the level of an individual, family, or wider system and are partly determined by the resources available. Although the importance of individual cognitive factors is evident from the literature, and thus was the focus of the current study, other factors clearly contribute to this complex and transactional process.

Ninety-two per cent of the variance in parental subjective wellbeing in this study was unaccounted for by the parental cognitive factors investigated. The theoretical models outlined (Lazarus & Folkman, 1984; Lazarus, 1991; McCubbin & Patterson, 1983; Wallander *et al.*, 1989) indicate that other variables that were not investigated in the current study (e.g. emotional, behavioural and social factors) could account for the remaining variance in parental subjective wellbeing and the pattern of results observed. For example, almost two thirds (63%) of the caregivers surveyed had other children at home. Parenting experiences with other children in the family are likely to influence these caregivers' perceptions of their role as parents and family functioning in general, thus the family context may account for the trend for most parents to be able to recognise positive gains of having a child with PMID. Furthermore, other factors implicated in family stress models, such as practical resources (e.g. finances, whether there are two caregivers in the family, services such as respite) are likely to contribute to the context within which caregivers' cognitive appraisals are formed.

For example, a single parent who is unable to work may feel more stressed and frustrated with the situation and thus may find it more challenging to identify positive gains of having a child with a disability. Thus it seems noteworthy that a high proportion of caregivers who were surveyed were married, in a civil partnership or were cohabiting, as this would indicate a protective factor. Given the main recruitment strategy of advertising the study via family support organisations, it is also likely that the families who took part in the study were able to access and make use of a range of supports and resources. This could explain the trend for most parents to be able to recognise positive gains of having a child with PMID in this study.

Subjective wellbeing of parents with PMID compared with other groups

It was hypothesised that self-reported subjective wellbeing would be lower in parents and family caregivers of children and young people with PMID than in parents of children with behavioural difficulties who do not have a disability, and the general population. This was based both on the theoretical models outlined (Lazarus & Folkman, 1984; Lazarus, 1991; McCubbin & Patterson, 1983; Wallander *et al.*, 1989) and on the findings of previous studies. Research suggests that increased daily stressors, care burden and higher levels of functional, intellectual or motor impairment are related to poorer parental psychological outcomes (Wallander & Varni, 1998), such as increased stress, anxiety, depression and poorer coping (e.g. Dunn *et al.*, 2001; Hassall *et al.*, 2005; Lloyd & Hastings, 2009; Lloyd & Hastings, 2009; Glenn *et al.*, 2009). The results of the current study supported this hypothesis, with the mean subjective wellbeing values of the sample of caregivers studied ($M= 39.39$; $SD= 7.57$) corresponding with the 11th percentile for subjective wellbeing in the general population (Bryson *et al.*, 2011).

This places parents of children with PMID alongside other groups experiencing low levels of support, autonomy, security and control in their lives or who are unemployed or experiencing levels of 'very bad' self-rated health (Bryson *et al.*, 2011). As well as subjective wellbeing being low in this group of caregivers in comparison with the general population, it was the equivalent of approximately the 25th percentile for pre-intervention subjective wellbeing in a group of parents taking part in a parenting early intervention programme (Lindsay *et al.*, 2008). This demonstrates that as a group, caregivers of children with PMID have much lower levels of wellbeing than the general population and lower levels of wellbeing than parents who have children with behavioural difficulties who do not have a disability. However, given the well-documented link between certain demographic indices (e.g. employment status, level of completed education) and health, quality of life and wellbeing (Bryson *et al.*, 2011), it is crucial to assess the findings of the current study within the wider context of these demographic and environmental factors (Olsson & Hwang, 2008). Emerson *et al.* (2006) urged researchers to consider the wider social context that families operate within, given that families of a child with a disability are at a greater risk

of exposure to socio-economic adversity and that wellbeing and psychological distress are associated with social position (Power, 2002).

Contextual and environmental factors

Family composition

With regard to the family composition, twenty per cent of caregivers of children with PMID in this study indicated that they were in a household with only one parental caregiver. This is comparable with 2011 national figures for the number of lone parents with dependent children in the UK (26%; Office for National Statistics, 2012). However, thirteen per cent of caregivers in the current study did not provide any response to the survey item that asked for details of those living within the family home, therefore it is likely that the twenty per cent figure is an underestimate. Parenting a child with PMID as a lone parent is likely to be more challenging than parenting in the context of a two parent household, simply due the fact that caregiving duties and burdens cannot be shared. One study found that single mothers were more likely to experience depression than their counterparts with partners (Blacher & Lopez, 1997) and if there had been a higher proportion of lone parents in this study, this itself might influence parental subjective wellbeing negatively.

Social networks and support

Neither social support nor characteristics of caregivers' social networks were explored in this study. However, social support is identified as a key resource that influences parental appraisals of the situation, coping behavior and outcomes in the key theoretical models in this area (Lazarus & Folkman, 1984; Lazarus, 1991; McCubbin & Patterson, 1983; Wallander *et al.*, 1989). Furthermore, several studies covered as part of the systematic review underlined the role of social support and the social context in the stress-coping process of having a child with a disability. This role is complex, and Hassall *et al.* (2005) suggested that family support facilitated the development of a more internal parental locus of control, which in turn is associated with more positive parent psychological outcomes. Social support also indirectly influenced parental adjustment and the reported severity of the child's symptoms via parental stress (Siman-Tov & Kaniel, 2011). Receiving social support seems to help parents feel more in control and encourages a sense of meaning, lowering parental stress (Siman-Tov & Kaniel, 2011). As such, social support could also have influenced levels of caregiver subjective wellbeing in this study.

Service provision

Even in the UK, where healthcare is free at the point of delivery via the National Health Service, there still exists geographical variation in support available to families of children with ID (Wright *et al.*, 2008). Service provision depends on policy priorities and service infrastructure, which vary across nations and even regions. A particular issue in the UK is

that of who has responsibility for service provision for children with ID and their families, as this differs across the devolved nations and even within them, depending on local authority and health board or trust agreements (Wright *et al.*, 2008). As the current study covered the whole of the UK and Ireland, service provision for families will have varied significantly depending on where they lived.

Whether a family feels well supported by services or not influences the context within which caregivers make appraisals about their situation. If those who participated were from areas with limited service provision (the survey did not ask about this), this is likely to impact on the family in terms of the care burden and associated demands they experience, and in terms of their general health and wellbeing due to cumulative stress and strain. These could have all impacted on the levels of caregiver wellbeing reported in this study.

Culture

Following on from this idea of geographical differences in service provision, Konstantareas, and Lampropoulou (1998) highlighted the influence of cultural norms, with regard to particular styles of family support, values and coping strategies employed across different cultures. It is important to remember the societal and cultural discourse contexts of families and caregivers as these are highly likely to influence cognitive appraisals of situations, their meaning and the accepted or expected ways of coping with stressors.

Parent demographics

In terms of parent demographic factors, there are several variables that may have influenced the caregiver subjective wellbeing scores reported in this study.

Employment

Fifty-two per cent of the sample was either in employment or studying and 48 per cent were not employed, the latter figure being much higher than the seasonally-adjusted national level of unemployment in the UK in April 2012: just over eight per cent of the labour market (Eurostat, 2012). A substantial body of evidence has reported that paid employment and job quality affect health and wellbeing (Bryson *et al.*, 2011) and it is likely that many parents in this group do not have the option of entering employment or their chosen career due to the restrictions of their caregiving duties. Therefore, this represents a further factor that could account for the pattern of decreased subjective wellbeing scores in the group of parents surveyed.

Health

In addition to restricted employment options, forty-five per cent of the sample reported that they had health issues themselves, including a number of chronic and serious conditions that impact on quality of life (see Table 6 for an overview). Many of these

conditions appeared to be stress-related and parental health status was significantly associated with child age in the current study. Parents who reported health issues tended to have children who were older and also identified fewer positive gains of having a child with PMID, with both of these associations reaching statistical significance (see Table 10). Therefore, caring for a child with PMID appears to result in a cumulative strain for caregivers that seems to affect their ability to identify positive aspects of caring for the child with PMID as time goes on. Nevertheless, it is not possible to infer the direction or nature of causality in these relationships, based on correlational analyses alone. In the general population, subjective wellbeing is highly associated with self-reported general health (Bryson *et al.*, 2011), which was a strong predictor of the wellbeing of parents who had children with ID in a study by Olsson and Hwang (2008). Thus the high percentage of health issues reported by this sample of parents of children with PMID may be a further explanatory factor accounting for the much lower subjective wellbeing scores seen in this group.

Education

Thirty per cent of the sample was educated to undergraduate level and a further nineteen per cent to postgraduate level, indicating that the sample were highly educated and possibly not representative of the wider population of parents of children with PMID. This suggests that the parental subjective wellbeing figures from the present study are likely to represent those in this particular population of parents that are functioning relatively well. Furthermore, these are the caregivers who are more likely to have had access to support networks such as peers and organisations providing family information, advice and support. Consequently, this study may have yielded observations of a higher level of wellbeing than may be the case in the remainder of this population, which is very concerning.

Clinical implications

The findings of this research study have implications for clinical practice, as practitioners often work closely with family members and caregivers as part of support provision for a child with PMID. The effect of parental locus of control on subjective wellbeing shows that how parents perceive and make sense of their situation is important, as families vary widely in what they consider challenging or not. Practitioner beliefs may be very different to those of caregivers', thus practitioners need to remember not to make assumptions based on their own attributions.

The anecdotal feedback provided by caregivers who participated in the study is of note to practitioners based in all settings. Some caregivers expressed that their own emotions and thoughts relating to being a parent of a significantly disabled and incredibly complex child and the challenges that brought were not often explored or acknowledged. This appears to be related to practitioners whose intentions are to avoid further upset or who do not feel

sufficiently skilled to explore such issues. Clinical psychologists are ideally placed to develop and deliver training for other practitioners who are less familiar with the psychological impacts of caring for a child with PMID. This can work well when clinical psychology input is embedded within a multi-disciplinary team and the clinical psychologist has the opportunity to work jointly and collaboratively with other professionals, whilst modelling and demonstrating the sorts of skills required to explore these issues with families.

Given the significant link between parental locus of control and wellbeing observed in the current study, it may be worth psychologically screening parents and other caregivers using brief measures of wellbeing, distress, parental locus of control or parental confidence. This would enable early identification of families who are likely to struggle, thus providing an opportunity for intervention and support before difficulties become salient at crisis. However, this clearly relies on the development and validation of psychometrically robust measures, whereas there are few available at the present time for parents of children with disabilities. Screening could be undertaken by staff such as nurses or paediatricians, with consultation and support from clinical psychology, and may be a good way of having difficult but important conversations.

Following on from the identification of caregiver psychological needs, a range of interventions need to be developed specifically for this group of parents. These could follow a 'stepped care' provision approach, whereby a selection of interventions of differing level of intensity and complexity are on offer and these are matched to the family needs at that point in time. At a basic level, staff training, consultation and joint working, as outlined above, are means of increasing other practitioners' psychological mindedness and skills. Provision of realistic advice and guidance, but at the right time when the family is ready for it, is another factor that seems likely to have an impact on parental cognitive appraisals, such as locus of control. CBT and self-help type interventions could then be offered one step up via clinical psychologists supervising other staff to take on these supervisory roles. Cognitive appraisals are clearly important, thus it makes sense to predict that existing interventions with a cognitive component may be of some use to some caregivers, depending on their individual and family circumstances and beliefs. Social support in a formal group setting from peers in similar situations may be a powerful way of normalising some caregivers' ideas about things and it also gives families the chance to check out some of their beliefs with other caregivers who understand the experience.

Some researchers in this area are advocating the use of acceptance and mindfulness based interventions for families of children with ID (MacDonald *et al.*, 2010). Interventions to help parents feel more empowered, capable and in control of parenting their child should be encouraged in whatever way possible, even if this is just over what seem like relatively

small things, such as praising them for their effort and determination. At present, the evidence base for interventions for parents of children with ID is almost non-existent. However, there does seem to be a converging pattern of findings emerging from the literature focused on parenting a child with ID. Practice-based research to evaluate any interventions that clinicians have found fruitful would be a good start to generating such an evidence base.

Ethical issues associated with online research

As the study was advertised using social media and made available via an online survey, issues relating to this research modality were considered. Online research is a relatively new methodology within the social sciences, but is now being used 'widely and with great enthusiasm' by psychologists (Reips, 2002). There are many benefits to conducting online psychological research; however, there are also several caveats that must be borne in mind.

Around 19 million UK households (77%) have an internet connection (Office of National Statistics, 2011), enabling many individuals to take part in research studies from the comfort of their own home. Presenting a study online also widens the net for recruitment of participants, potentially increasing diversity within the sample and thus improving generalisability. The systematic review identified that fathers were underrepresented in parent research, thus online recruitment and presentation of the current study was one means of attempting to make it more accessible and appealing to fathers. Increased accessibility also encourages sample heterogeneity as participants are more likely to be spread out over a wider geographical area (Gardner, 2007). Increased accessibility also means that the study can be accessed round the clock, so individuals can participate at a time that is convenient for them. This issue was particularly pertinent to the target population of this study, given their significant caring commitments. Online survey data can also automatically be stored, thus minimising the opportunity for error associated with manual data entry.

Kraut *et al.* (2007) highlight, however, that generalising from internet samples can be problematic, given the demographic differences in internet users. In 2006, only twenty-one per cent of 'low-income' households in the UK had internet access, compared with ninety-four per cent of 'high-income' households (Office for National Statistics, 2006). This suggests that internet based recruitment may yield unrepresentative samples. Gardner (2007) echoes this sentiment, stating that the majority of internet users are white, middle-class males. Furthermore, the environment of participants cannot be controlled when completing the study online. Factors such as noise, interruptions and competing priorities may confound the results or even lead to non-completion. Technical difficulties can also perturb participants and may lead to abandonment of the study.

Although considered no more risky than traditional psychological research methods (Kraut *et al.*, 2007), online research poses some additional ethical issues due to the nature of the research context. The absence of direct contact with participants makes it difficult to monitor and respond to any distress or lack of understanding during the study. In addition, the issue of informed consent must be considered carefully in online research because the researcher is unable to gauge the participant's understanding, as they are able to do in person. One strategy for overcoming this is to break up the information provided prior to giving consent into smaller sections (Gardner, 2007; Kraut *et al.*, 2007). The BPS (2007) also suggests that contact details of the researcher are clearly given in case an individual would like to clarify something or withdraw from the experiment. A debriefing should also be provided, ideally appearing automatically if the participant closes the experiment window (Kraut *et al.*, 2007).

All of these considerations were taken into account prior to undertaking the current study and it was decided that an online survey remained an appropriate tool as it would offer the opportunity to recruit more participants, especially fathers, from a group that is underrepresented in the research literature (Olsson & Hwang, 2008). All of the questionnaires presented online had been used previously in peer-reviewed research and were deemed acceptable by both the School of Health in Social Science Ethics Committee and the organisations who reviewed the study prior to advertising it. All information was presented on web pages as clearly and simply as possible and participants were only able to proceed to the online questionnaire items if they had clicked the 'accept' button that constituted giving their informed consent. A debriefing was provided at the end of the study and contact details of the author were also given. Furthermore, the author invited each organisation to direct any feedback relating to participation in the study to her in order to address any issues arising for participants. This was acknowledged in the summary provided to all organisations involved in recruitment to be shared with participants.

Strengths and limitations of the study

This is the first known study to investigate the subjective wellbeing of parents of children with PMID. Useful demographic information was gathered about this population that can be used to make comparisons with other groups. For example, it was particularly noteworthy that such a high proportion of parents had health issues themselves in the group surveyed. These observations of group characteristics would form an ideal basis for future studies, as it would be informative to understand the specific issues faced by this population of parents and elucidate causal mechanisms and common trajectories in order to inform support provided. Furthermore, the current study offers a number of insights that are worthy of further exploration in this population and several suggestions for future research are outlined in the following section.

It is hoped that this study generates some interest and momentum for further research in this area. In addition to the applications of the study for research purposes, a number of clinical implications were also outlined, thus a further strength of the study is the applicability of the findings to an applied context. Nevertheless, the study also had a number of limitations which can be broadly grouped into the categories of sampling, measurement, design, and confounding variables. This must be taken into consideration alongside the findings, limiting their generalisation.

Sampling

There is not currently a single definition of PMID that is accepted globally and a multitude of labels exist that are used to describe this group (Nakken & Vlaskamp, 2007; Pawlyn & Carnaby, 2009). These often overlap with other groups who, for example do not have ID (e.g. children with complex/high support needs; children who are technology dependent; medically fragile children). Therefore it can be difficult both in the literature and in practice to know exactly who is being referred to (Nakken & Vlaskamp, 2007). The definition of PMID adopted in the current study was selected on the basis of it capturing the multiple physical disabilities and also the profound ID of these children, in combination with their fragile health and their functioning at the very early stages of development (Pawlyn & Carnaby, 2009). This is also in line with clinical descriptions used in practice in ID services that characterise specific aspects of the child's functioning that are limited, thus giving an idea of the type of service needs of families (Pawlyn & Carnaby, 2009).

However, there remains a wider issue of the challenge of defining this group that contributes to the lack of universal terminology used. Nakken and Vlaskamp (2007) highlight the fact that this group of individuals has such profound ID that existing standardised tools for assessing intellectual functioning cannot offer a valid estimation of their intellectual capacity. This is because they have little to no apparent understanding of verbal language or symbolic interactions with objects and have no ability for self-support (Hogg & Sebba, 1986). Thus, there is a real need for assessment tools that are sensitive enough to confirm or disconfirm such impaired levels of functioning and that would both inform practice and enable clearer delineation of this population from other similar populations in research (Nakken & Vlaskamp, 2007). In the context of the present study, parents self-selected based on the definition of PMID provided and it was not possible to verify whether their child met these criteria or not. However, this is a common selection method in ID research (Emerson *et al.*, 2006) and given the wider issues of defining this group in research and practice, there were no obvious alternatives.

A separate limitation of this study in relation to sampling concerns whether the group of parents surveyed was representative enough of the population of UK parents of children with PMID to generalise the findings more widely beyond the sample surveyed. However,

there is little information available in the literature about the demographic characteristics of this group available for comparison. Although efforts were made to encourage participation of fathers in this population, they still made up a very small proportion (3%) of the caregiver sample studied. This may be due to fathers not identifying themselves as 'caregivers' or it might be the case that mothers were more involved with the parent organisations that aided in the recruitment of participants. However, it is a significant limitation, as there are documented gender differences in the way mothers and fathers make sense of and adjust to parenting a child with additional needs (Hastings *et al.*, 2005a; Hastings *et al.*, 2005b; Janssens, 1994; Lanfranchi & Vianello, 2012)

An additional feature of this sample that limits generalisability is the fact that ninety-seven per cent of participants and ninety-four per cent of their children with PMID were of white ethnicity (see Tables 5 and 7). This figure is higher than those that have been quoted regarding the ethnicity of children with and without ID in the British population as a whole (90% and 91% white ethnicity respectively; Emerson & Hatton, 2007; Meltzer *et al.*, 2000). Emerson and Hatton (1999) used 1991 Census data and population projections and estimated that the non-White UK population will have increased from five per cent in 1991 to eight per cent in 2021. There is evidence that families with a child with ID from minority ethnic groups consistently face disadvantages (e.g. financial) when compared with families with a child with ID from majority ethnic communities, and when compared to others from the same ethnic group (Hatton, 2002). These families report high levels of need for themselves and their children with ID, which are closely linked with a lack of both formal and informal support systems (Hatton, 2002). Therefore, again, it is likely that as the sample studied included a high proportion of caregivers who were white, the current findings represent those who are more advantaged within this group and thus may not represent the experiences of the wider population, including families from other ethnic backgrounds.

Measurement

As noted in the systematic review, the measure of parental locus of control employed in the current study (PLOC-R) has some limitations in terms of its psychometric properties. Several adaptations of the original measure are currently in use and so it is difficult to compare findings across studies that use this measure (Lloyd & Hastings, 2009). It has been reported that the PLOC would benefit from more use with larger samples to ensure reliability (Hagekull *et al.*, 2001). In addition, the discriminant validity of the PLOC requires further investigation, as it could be measuring other constructs such as parental distress or other cognitive variables (Lloyd & Hastings, 2009). However, this measure was selected for use in the current study as there is currently no psychometrically sound instrument available to measure parental locus of control that is developed specifically for or is well validated with caregivers of children with intellectual disability. Other tools are available

that measure this and similar constructs, such as parental self-efficacy, self-esteem, perceived competence and parental satisfaction. However, they suffer similar limitations. The issues of social desirability effects and the limitations of the PGS have already been discussed and so will not be repeated here.

A limitation identified by caregivers themselves in the feedback they provided was that the quantitative questionnaires used did not seem like they were developed to assess the specific experiences of those caring for a child with PMID. Some caregivers expressed that the measures failed to capture the content and richness of their experiences and they made suggestions that future research should explore the emotional impact of having a child with PMID and how confident and supported caregivers feel in their role. These questions are similar to those included in the Sheffield Learning Disability Outcome Measure (SLDOM; Sheffield Children's NHS Foundation Trust, n.d.) that was considered in the initial design stages of the current study. The SLDOM is also used routinely as an outcome measure in clinical practice in child learning disability services. However, although it is freely available online, this measure is unpublished and has not been subject to any evaluation of its psychometric properties. Therefore it was not selected for use in the present study.

Design

The cross-sectional design of the study limited the ability to explore causal and temporal relationships, although this is a limitation of much of the research on parental locus of control in this and similar populations (Siman-Tov & Kaniel, 2011). In this case, this was due to limited resources and time constraints, but it would be useful to replicate the study using a longitudinal design, as beliefs related to parental locus of control may also interact with other coping resources and previous coping experiences to influence parent psychological outcomes (Beresford, 1994; Hassall *et al.*, 2005).

Several studies discussed in the systematic review undertook analyses relating to the five subscales of the PLOC (Parental Efficacy, Parental Responsibility, Child Control of Parent's Life, Parental Belief in Fate/Chance and Parental Control of Child's Behaviour), in addition to analyses using the total score for this measure. The Child Control and Parent Control subscales strongly correlated with parental stress (Hassall *et al.*, 2005), as was the case in previous research (Campis *et al.*, 1986; Hagekull *et al.*, 2001). Lloyd and Hastings (2009) similarly found that the Parental Control subscale correlated significantly with all maternal wellbeing measures used in their study and significantly predicted positive perceptions and anxiety at time one. Therefore, analyses of these subscales in the present study would have added value as these two subscales show the most utility in predicting parenting stress.

Other studies have used median-split analyses to explore characteristics associated with subgroups of samples of parents (e.g. using low/high wellbeing group comparisons). This

would have been informative as it would have enabled fuller exploration of the characteristics of caregivers with high levels of wellbeing. A final weakness of the current study in terms of its design was the lack of a comparison group to control for the influence of other variables, as this would have enabled clearer delineation of the effects observed.

With regard to the limitations of the overall aims and objectives of the current study, sufficient time and resources were not available to explore the full range factors that have been identified in the theoretical models of the stress-coping process. Therefore the study focused on two specific cognitive factors identified as pertinent in the systematic review and wider literature. It is unsurprising therefore that the only statistically significant predictor to emerge from the inferential analyses (parental locus of control) accounted for just eight per cent of the variance in parental subjective wellbeing scores for this group. Clearly, the issues influencing subjective wellbeing in this group are complex and go beyond cognitive factors and further research is needed to test the various theoretical models outlined in full with this population of parents.

Directions for future research

A number of limitations of the present study have been discussed in depth, with reference to how future research studies might seek to address some of these limitations. In addition to this, the current study raised a number of further questions that would merit further investigation and would add to the existing literature.

First, it would be interesting to explore whether there are any relationships between parental locus of control, perceived child behaviour problems and parental sense of competency in caregivers of children with PMID. The research reviewed suggested that these were important factors in other studies of a broader population of parents of children with ID. It is likely that there is variation within this group of families in terms of child characteristics (e.g. current health status/prognosis, child behaviours, temperament) so it would also be useful to investigate whether these are related to caregiver subjective wellbeing. Furthermore, it would be useful to investigate whether locus of control influences subjective wellbeing and other psychological outcomes in paid caregivers and other staff working with children with PMID in caring roles.

The conceptual overlap between parental locus of control and related concepts such as parental self-efficacy, parent self-esteem, perceived competence and parental sense of coherence needs to be investigated further in order to delineate the contribution of each and further distinguish these from one another so that they represent useful concepts for research and practice.

The importance of relational variables such as relationship quality, attachment styles and social support emerged in the current study, although they were not investigated. Future research exploring whether these relational variables are associated with parental cognitions would be valuable to our understanding of stress and coping within this group, particularly with regard to the identification of protective factors. Along these lines, further research could build on the findings of the current study in order to work out whether the observed pattern of positive gains in this sample could be explained with reference to the family context and experiences. The trend for most parents to be able to identify positive gains of having a child with PMID may actually reflect parental positive perceptions of themselves as a parent rather than of their child. The presence of other children in the household may also influence caregivers' ability to recognise positive gains of having a child with a disability, as they have alternative experiences of caregiving and parenting as a point of comparison.

A final suggestion for future research is the need for qualitative explorations of the experience of parents of children with PMID in order to gain an insight into their lived experiences and the emotional impact of caring for a very fragile child. Explorations in relation to caregivers' experiences over time would be incredibly informative, paying attention to caregiver and family experiences in terms of where the child with PMID is at developmentally, where the family is in their family life cycle and the stage in the course of the disease or disability (Wallander & Varni, 1998). Qualitative research could also explore caregivers' opinions on what they believe would be helpful or useful in relation to their subjective wellbeing.

Summary and conclusions

The present study demonstrated that a more external parental locus of control was related to lower levels of parental subjective wellbeing in caregivers of children with PMID. Furthermore, regression analysis revealed that, as hypothesised, parental locus of control significantly predicted the subjective wellbeing of parents of children with PMID and accounted for around eight per cent of the variance in parental subjective wellbeing. The mean parental subjective wellbeing value for caregivers of children with PMID in this study corresponded with the 11th percentile for subjective wellbeing in the general population (Bryson *et al.*, 2011), indicating low levels of subjective wellbeing in this group. These findings are line with previous research highlighting a relationship between parental locus of control and negative parent psychological outcomes (Dunn *et al.*, 2001; Glenn *et al.*, 2009; Hassall *et al.*, 2005; Lloyd & Hastings, 2009). Furthermore, this is the first known study to investigate the relationship between these variables in this specific subgroup of parents of children with ID who have a very complex profile of physical and psychological needs.

With regard to implications for clinical practice and service provision, the effect of parental locus of control on subjective wellbeing shows that how parents perceive and make sense of their situation is important, as families vary widely in what they consider challenging or not. Thus practitioners need to remember not to make assumptions based on their own attributions. Caregivers who took part in the study expressed dissatisfaction regarding practitioners' lack of attention to the emotional impact of caring for a child with PMID. Clinical psychologists are well-placed to address potential staff training and awareness issues that may account for this, in addition to providing supervision and consultation to encourage consideration of the psychological aspects of families' experiences. The usefulness of screening caregivers of children with PMID for psychological distress was discussed in relation to early identification and support that may promote better outcomes. However, the evidence base for interventions that facilitate positive psychological outcomes for this group of parents is extremely limited and this urgently needs to be developed, perhaps starting with practice-based evidence.

Strengths of the study were that this is the first known study to investigate the subjective wellbeing of parents of children with PMID. Observations of characteristics of this group would form an ideal basis for future studies and a number of insights are reported that are worthy of further exploration in this population. A further strength of the study is the applicability of the findings to an applied context. A number of limitations of the present study were discussed in depth, covering the domains of sampling, measurement, design, and confounding variables. Suggestions for further research that either overcomes these methodological limitations or builds on some of the interesting insights have been provided and it this will hopefully result in further interest and research in this area for the benefit of these families.

REFERENCES

- American Psychological Association (2009). *Publication manual of the American Psychological Association (6th Edn.)*. Washington, DC: Author.
- Baker B.L., Blacher J. & Olsson M.B. (2005). Preschool children with and without developmental delay: behaviour problems, parents' optimism and well-being. *Journal of Intellectual Disability Research*, 49, 575-590.
- Bandura A. (1982). Self-efficacy mechanism in human agency. *American Psychologist*, 37, 122-147.
- Beresford B.A. (1994) Resources and strategies: How parents cope with the care of a disabled child. *Journal of Child Psychology and Psychiatry*, 35, 171-209.
- Blacher, J. & Lopez, S. (1997) Contributions to depression in Latina mothers with and without children with retardation: implications for caregiving. *Family Relations: Interdisciplinary Journal of Applied Family Studies*, 46, 325-334.
- Brace, N., Kemp, R. & Snelgar, R. (2009). *SPSS for psychologists (4th edn)*. London: Palgrave Macmillan.
- British Psychological Society (2004). *Editorial style guide*. Leicester: Author.
- British Psychological Society. (2007). *Code of ethics and conduct*. Leicester: British Psychological Society.
- Bryson, A., Green, F., Bridges, S. & Craig, R. (2011). Health survey for England 2010: Well-being, health and work. Retrieved 28 June 2012, from: ic.nhs.uk/webfiles/publications/003_Health_Lifestyles/HSE2010_REPORT/HSE2010_Ch7_Well_being_health_and_work.pdf
- Bytheway, B. & Johnson, J. (2008). *The social construction of carers*. In J. Johnson & C. De Souza (eds.) *Understanding health and social care*. London: Sage.
- Campis L.K., Lyman R.D., & Prentice-Dunn, S. (1986). The Parental Locus of Control Scale: Development and validation. *Journal of Clinical Child Psychology*, 15, 260-267.
- Diener, E. (1984). Subjective well-being. *Psychological Bulletin*, 95, 542-575.
- Dunn, M.E., Burbine, T., Bowers, C.A., & Tantleff-Dunn, S. (2001). Moderators of stress in parents of children with autism. *Community Mental Health Journal*, 37, 39-52.
- Emerson, E. & Hatton, C. (2007). Mental health of children and adolescents with intellectual disabilities in Britain. *British Journal of Psychiatry*, 191, 493-499.
- Emerson, E. & Hatton, C. (1999). Future trends in the ethnic composition of British society and among British citizens with learning disabilities. *Tizard Learning Disability Review*, 4, 28-32.
- Emerson, E., Hatton, C., Llewellyn, G., Blacher, J., & Graham, H. (2006). Socioeconomic position, household composition, health status and indicators of the well-being of mothers of children with and without intellectual disabilities. *Journal of Intellectual Disability Research*, 50, 862-73.
- Eurostat (2012). Unemployment rate (UK): Seasonally adjusted data. Retrieved 31 July 2012 from: www.google.co.uk/publicdata

- Faul, F., Erdfelder, E., Buchner, A. & Lang, A.-G. (2007). Statistical power analyses using G*Power 3.1: Tests for correlation and regression analyses. *Behavior Research Methods*, 41, 1149-1160.
- Faul, F., Erdfelder, E., Lang, A.-G. & Buchner, A. (2009). G*Power 3: A flexible statistical power analysis program for the behavioral, and biomedical sciences. *Behavior Research Methods*, 39, 165-191.
- Field, A.P. (2009). *Discovering statistics using SPSS* (3rd edn). London: Sage.
- Fitton, P. (1994). *Listen to me: Communicating the needs of people with profound and multiple disabilities*. London: Jessica Kingsley.
- Glenn, S., Cunningham, C., Poole, H., Reeves, D., & Weindling, M. (2009). Maternal parenting stress and its correlates in families with a young child with with cerebral palsy. *Child: Care, Health and Development*, 35(1), 71-78.
- Friedrich, W.N., Cohen, D.S. & Wiltner, L.T. (1988). Specific beliefs as moderator variables in maternal coping with mental retardation. *Children's Health Care*, 17(1), 40-44.
- Gardner, K. (2007). Conducting your PhD via the internet. *The Psychologist*, 20(4), 232-233.
- Hagekull, B., Bohlin, G., & Hammarberg, A. (2001) The role of parental perceived control in child development: a longitudinal study. *International Journal of Behavioral Development*, 25, 429-437.
- Hassall, R., Rose, J., & McDonald, J. (2005). Parenting stress in mothers of children with an intellectual disability: The effects of parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research*, 49, 405-418.
- Hastings, R.P., & Johnson E. (2001). Stress in UK families conducting intensive home-based behavioral intervention for their young child with autism. *Journal of Autism and Developmental Disorders*, 31, 327-336.
- Hastings R. P. & Taunt H. M. (2002). Positive perceptions in families of children with developmental disabilities. *American Journal on Mental Retardation*, 107, 116-127.
- Hastings R.P. & Brown T. (2002). Behavior problems of children with autism, parental self-efficacy, and mental health. *American Journal on Mental Retardation* 107, 222 - 232.
- Hastings, R.P., Allen, R., McDermott, K., & Still, D. (2002). Factors related to positive perceptions in mothers of children with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities*, 15, 269-275.
- Hastings, R.P., Beck, A., & Hill, C. (2005). Positive contributions made by children with an intellectual disability in the family: Mothers' and fathers' perceptions. *Journal of Intellectual Disabilities*, 9, 155-165.
- Hastings, R P., Kovshoff, H., Brown, T., Ward, N.J., Espinosa, F.D. & Remington, B. (2005). Coping strategies in mothers and fathers of preschool and school-age children with autism. *Autism*, 9, 377-391.

- Hatton, C. (2002). People with intellectual disabilities from ethnic minority communities in the United States and United Kingdom. In: L. Masters Glidden (Ed.) *International Review of Research in Mental Retardation*, 25 (pp. 209-239). San Diego: Elsevier Academic Press .
- Heiman, T. (2002). Parents of children with disabilities: Resilience, coping and future expectations. *Journal of Developmental and Physical Disabilities*, 14(2), 159-171.
- Hogg, J. & Sebba, J. (1986). *Profound retardation and multiple impairment*. London: Croom-Helm.
- Kaplan, A. (2008). Clarifying meta-cognition, self-regulation, and self-regulated learning: What's the purpose? *Educational Psychology Review*, 20(4), 477-484.
- James, W. H., & Rotter, J.B. (1958). Partial and 100% reinforcement under chance and skill conditions. *Journal of Experimental Psychology*, 55, 397-403.
- Janssens, J.M.A.M. (1994). Authoritarian child rearing, parental locus of control, and the child's behaviour style. *International Journal of Behavioral Development*, 17(3), 485-501.
- Kingsley, E.P. (1987). *Welcome to Holland*. Retrieved 23 November 2011 from our-kids.org/Archives/Holland.html
- Konstantareas, M. M., & Lampropoulou, V. (1998). Stress in Greek mothers with deaf children: Effects of child characteristics, family resources and cognitive set. *American Annals of the Deaf*, 140(3), 264-270.
- Kraut, R., Olson, J., Banaji, Bruckman, A., Cohen, J., & Couper, M. (2007). Psychological Research Online. *American Psychologist*, 59, 105-117.
- Lanfranchi, S. & Vianello, R. (2012). Stress, locus of control and family cohesion and adaptability in parents of children with Down, Williams, Fragile X, and Prader-Willi syndromes. *American Journal on Intellectual and Developmental Disabilities*, 117(3), 207-224.
- Lazarus, R.S. (1991). *Emotion and adaptation*. New York, NY: Oxford University Press.
- Lazarus, R.S. & Folkman, S. (1984). *Stress, appraisal, and coping*. New York, NY: Springer.
- Lefcourt, H.M. (1982). *Locus of control: Current trends in theory and research*. Hillsdale, NJ: Lawrence Erlbaum Associates.
- Lindsay, G., Davies, H., Band, S., Cullen, M.A., Cullen, S., Strand, S., Hasluck, C., Evans, R. & Stewart-Brown, S. (2008). *Parenting early intervention pathfinder evaluation*. Department for Children, Schools and Families.
- Lloyd, T. & Hastings, R.P. (2008). Psychological variables as correlates of adjustment in mothers of children with intellectual disabilities: Cross-sectional and longitudinal relationships. *Journal of Intellectual Disability Research*, 52, 37-48.
- Lloyd, T. & Hastings, R.P. (2009a). Parental locus of control and psychological well-being in mothers of children with intellectual disability. *Journal of Intellectual and Developmental Disability*. 34(2), 104-115.

- Lloyd, T. & Hastings, R.P. (2009b). Hope as a psychological resilience factor in mothers and fathers of children with intellectual disabilities. *Journal of Intellectual Disability Research*, 53(12), 957-968.
- MacDonald, E.E., Hastings, R.P. & Fitzsimons, E. (2010). Psychological acceptance mediates the impact of the behaviour problems of children with intellectual disability on fathers' psychological adjustment. *Journal of Applied Research in Intellectual Disabilities*, 23(1), 27-37.
- McCubbin H.I. & Patterson J.M. (1983). The family stress process: The Double ABCX Model of adjustment and adaptation. In: H.I. McCubbin, M.B. Sussman & J.M. Patterson (eds), *Social Stress and the Family: Advances and Developments in Family Stress Theory and Research*, pp. 7-37. New York, NY: Haworth Press.
- McKinney, B., & Peterson, R.A. (1987). Predictors of stress in parents of developmentally disabled children. *Journal of Pediatric Psychology*, 12(1), 133-150.
- Meltzer, H., Gatward, R., Goodman, R. et al. (2000). *Mental health of children and adolescents in Great Britain*. London: The Stationery Office.
- Mencap & PMLD Network (n.d.). European Union (n.d.). *About profound and multiple learning disabilities*. Retrieved 10 June 2011 from: pmldnetwork.org/PMLD%20Definition%20factsheet%20-%20standard.pdf
- Miller A. C., Gordon R. M., Daniele R. J., & Diller L. (2001). Stress, appraisal, and coping in mothers of disabled and nondisabled children. *Journal of Pediatric Psychology*, 17, 587-605.
- Nakken, H. & Vlaskamp, C. (2007). A need for a taxonomy for profound intellectual and multiple disabilities. *Journal of Policy and Practice in Intellectual Disabilities*, 4(2), 83-87.
- Office for National Statistics (2012). Lone parents with dependent children. Retrieved June 28, 2012 from: www.ons.gov.uk/ons/dcp171780_251303.pdf
- Office for National Statistics (2011). *Statistical bulletin. Internet access: Households and individuals 2011*. Retrieved June 28, 2012 from: www.ons.gov.uk/ons/dcp171778_227158.pdf
- Olsson, M. B., & Hwang, C. P. (2008). Socioeconomic and psychological variables as risk and protective factors for parental well-being in families of children with intellectual disabilities. *Journal of Intellectual Disability Research*, 52(12), 1102-1113.
- Pawlyn, J., & Carnaby, S. (2009). *Profound intellectual and multiple disabilities: Nursing complex needs*. Chichester: John Wiley and Sons.
- Pit-ten Cate, I. (2003). *Family adjustment to disability and chronic illness in children*. University of Southampton: Unpublished PhD thesis.
- Power, C., Stansfeld, S.A., Matthews, S., Manor, O. & Hope, S. (2002) Childhood and adulthood risk factors for socio- economic differentials in psychological distress: Evidence from the 1958 British birth cohort. *Social Science and Medicine*, 55, 1989-2004.

- Rapanaro, C., Bartu, A. & Lee A.H. (2008). Perceived benefits and negative impact of challenges encountered in caring for young adults with intellectual disabilities in the transition to adulthood. *Journal of Applied Research in Intellectual Disabilities*, 21, 34-37.
- Reips, U.D. (2002). Standards for Internet-based experimenting. *Experimental Psychology*, 49(4), 243-256.
- Robinson, E.A. & Anderson, L.L. (1983). Family adjustment, parental attitudes and social desirability. *Journal of Abnormal Child Psychology*, 11(2), 247-256.
- Rotter, J.B. (1954). *Social learning and clinical psychology*. New York: Prentice-Hall.
- Rotter, J.B. (1966). Generalized expectancies for internal versus external control of reinforcements. *Psychological Monographs*, 80, Whole No. 609.
- Rotter, J.B. (1975). Some problems and misconceptions related to the construct of internal versus external control of reinforcement. *Journal of Consulting and Clinical Psychology*, 43, 56-67.
- Ruini, C., Ottolini, F., Rafanelli, C., Tossani, E., Ryff, C.D., Fava, G.A. (2003). The relationship of psychological well-being to distress and personality. *Psychotherapy and Psychosomatics*, 72(5), 268-275.
- Seligman, M.E.P. & Csikszentmihalyi M. (2000). Positive psychology: An introduction. *American Psychologist*, 55, 5-14.
- Sheffield Children's NHS Foundation Trust (n.d.). *Sheffield Learning Disabilities Outcome Measure*. Retrieved 10 June, 2011 from: www.corc.uk.net/resources/measures/parent/
- Siman-Tov, A., & Kaniel, S. (2011). Stress and personal resource as predictors of the adjustment of parents to autistic children: A multivariate model. *Journal of Autism and Developmental Disorders*, 41, 879-890.
- Singh, N.N., Lancioni, G.E., Winton, A.S.W., Fisher, B.C., Wahler, R.G., McAleavey, K. et al. (2006). Mindful parenting decreases aggression, noncompliance, and self-injury in children with autism. *Journal of Emotional and Behavioral Disorders*, 14, 169-177.
- Stewart-Brown, S., Tennant, A., Tennant, R., Platt, S., Parkinson, J. & Weich, S. (2009). Internal construct validity of the Warwick-Edinburgh Mental Well-being Scale (WEMWBS): A Rasch analysis using data from the Scottish Health Education Population Survey. *Health and Quality of Life Outcomes*, 7(15). Retrieved 10 June 2011 from hqlo.com/content/7/1/15
- Tennant, R., Fishwick, R., Platt, S., Joseph, S., & Stewart-Brown, S. (2007). *Monitoring positive mental health in Scotland: Validating the Affectometer 2 scale and developing the Warwick Edinburgh Mental Well-being Scale for the UK*. Edinburgh: NHS Health Scotland.
- Wallander, J.L., & Varni, J.W. (1998). Effects of pediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry*, 39(1), 29-46.

- Wallander, J.L., Varni, J.W., Babani, L., DeHaan, C.B., Wilcox, K.T., & Banis, H.T. (1989). The social environment and the adaptation of mothers of physically handicapped children. *Journal of Pediatric Psychology*, 14, 371-387.
- Weiner, B. (2006). *Social motivation, justice, and the moral emotions: An attributional approach*. New Jersey, NJ: Lawrence Erlbaum Associates.
- Wright, B., Williams, C. & Richardson, G. (2008). Services for children with learning disabilities. *The Psychiatrist*, 32, 81-84.

APPENDIX 1: Author guidelines for the journal Research in Developmental Disabilities

Preparation

Use of word processing software

It is important that the file be saved in the native format of the word processor used. The text should be in single-column format. Keep the layout of the text as simple as possible. Most formatting codes will be removed and replaced on processing the article. In particular, do not use the word processor's options to justify text or to hyphenate words. However, do use bold face, italics, subscripts, superscripts etc. When preparing tables, if you are using a table grid, use only one grid for each individual table and not a grid for each row. If no grid is used, use tabs, not spaces, to align columns. The electronic text should be prepared in a way very similar to that of conventional manuscripts (see also the Guide to Publishing with Elsevier: <http://www.elsevier.com/guidepublication>). Note that source files of figures, tables and text graphics will be required whether or not you embed your figures in the text. See also the section on Electronic artwork. To avoid unnecessary errors you are strongly advised to use the 'spell-check' and 'grammar-check' functions of your word processor.

Article structure

Subdivision - numbered sections

Divide your article into clearly defined and numbered sections. Subsections should be numbered 1.1 (then 1.1.1, 1.1.2, ...), 1.2, etc. (the abstract is not included in section numbering). Use this numbering also for internal cross-referencing: do not just refer to 'the text'. Any subsection may be given a brief heading. Each heading should appear on its own separate line.

Introduction

State the objectives of the work and provide an adequate background, avoiding a detailed literature survey or a summary of the results.

Material and methods

Provide sufficient detail to allow the work to be reproduced. Methods already published should be indicated by a reference: only relevant modifications should be described.

Theory/calculation

A Theory section should extend, not repeat, the background to the article already dealt with in the Introduction and lay the foundation for further work. In contrast, a Calculation section represents a practical development from a theoretical basis.

Results

Results should be clear and concise.

Discussion

This should explore the significance of the results of the work, not repeat them. A combined Results and Discussion section is often appropriate. Avoid extensive citations and discussion of published literature.

Conclusions

The main conclusions of the study may be presented in a short Conclusions section, which may stand alone or form a subsection of a Discussion or Results and Discussion section.

Appendices

If there is more than one appendix, they should be identified as A, B, etc. Formulae and equations in appendices should be given separate numbering: Eq. (A.1), Eq. (A.2), etc.; in a subsequent appendix, Eq. (B.1) and so on. Similarly for tables and figures: Table A.1; Fig. A.1, etc.

Essential title page information

- Title. Concise and informative. Titles are often used in information-retrieval systems. Avoid abbreviations and formulae where possible.
- Author names and affiliations. Where the family name may be ambiguous (e.g., a double name), please indicate this clearly. Present the authors' affiliation addresses (where the actual work was done) below the names. Indicate all affiliations with a lower-case superscript letter immediately after the author's name and in front of the appropriate address. Provide the full postal address of each affiliation, including the country name and, if available, the e-mail address of each author.
- Corresponding author. Clearly indicate who will handle correspondence at all stages of refereeing and publication, also post-publication. Ensure that telephone and fax numbers (with country and area code) are provided in addition to the e-mail address and the complete postal address. Contact details must be kept up to date by the corresponding author.
- Present/permanent address. If an author has moved since the work described in the article was done, or was visiting at the time, a 'Present address' (or 'Permanent address') may be indicated as a footnote to that author's name. The address at which the author actually did the work must be retained as the main, affiliation address. Superscript Arabic numerals are used for such footnotes.

Abstract

A concise and factual abstract is required. The abstract should state briefly the purpose of the research, the principal results and major conclusions. An abstract is often presented separately from the article, so it must be able to stand alone. For this reason, References should be avoided, but if essential, then cite the author(s) and year(s). Also, non-standard

or uncommon abbreviations should be avoided, but if essential they must be defined at their first mention in the abstract itself.

Graphical abstract

A Graphical abstract is optional and should summarize the contents of the article in a concise, pictorial form designed to capture the attention of a wide readership online. Authors must provide images that clearly represent the work described in the article. Graphical abstracts should be submitted as a separate file in the online submission system. Image size: Please provide an image with a minimum of 531 × 1328 pixels (h × w) or proportionally more. The image should be readable at a size of 5 × 13 cm using a regular screen resolution of 96 dpi. Preferred file types: TIFF, EPS, PDF or MS Office files. See <http://www.elsevier.com/graphicalabstracts> for examples.

Authors can make use of Elsevier's Illustration and Enhancement service to ensure the best presentation of their images also in accordance with all technical requirements.

Highlights

Highlights are mandatory for this journal. They consist of a short collection of bullet points that convey the core findings of the article and should be submitted in a separate file in the online submission system. Please use 'Highlights' in the file name and include 3 to 5 bullet points (maximum 85 characters, including spaces, per bullet point). See <http://www.elsevier.com/highlights> for examples.

Keywords

Abbreviations should be held to a minimum and should appear only after the full length term has been spelled out once in the text.

Acknowledgements

Collate acknowledgements in a separate section at the end of the article before the references and do not, therefore, include them on the title page, as a footnote to the title or otherwise. List here those individuals who provided help during the research (e.g., providing language help, writing assistance or proof reading the article, etc.).

Math formulae

Present simple formulae in the line of normal text where possible and use the solidus (/) instead of a horizontal line for small fractional terms, e.g., X/Y. In principle, variables are to be presented in italics. Powers of e are often more conveniently denoted by exp. Number consecutively any equations that have to be displayed separately from the text (if referred to explicitly in the text).

Footnotes

Footnotes should be used sparingly. Number them consecutively throughout the article, using superscript Arabic numbers. Many word processors build footnotes into the text, and this feature may be used. Should this not be the case, indicate the position of footnotes in the text and present the footnotes themselves separately at the end of the article. Do not include footnotes in the Reference list.

Table footnotes

Indicate each footnote in a table with a superscript lowercase letter.

Artwork

Electronic artwork General points

- Make sure you use uniform lettering and sizing of your original artwork.
- Save text in illustrations as 'graphics' or enclose the font.
- Only use the following fonts in your illustrations: Arial, Courier, Times, Symbol.
- Number the illustrations according to their sequence in the text.
- Use a logical naming convention for your artwork files.
- Provide captions to illustrations separately.
- Produce images near to the desired size of the printed version.
- Submit each figure as a separate file.

A detailed guide on electronic artwork is available on our website:

<http://www.elsevier.com/artworkinstructions>

You are urged to visit this site; some excerpts from the detailed information are given here.

Formats

Regardless of the application used, when your electronic artwork is finalised, please 'save as' or convert the images to one of the following formats (note the resolution requirements for line drawings, halftones, and line/halftone combinations given below): EPS: Vector drawings. Embed the font or save the text as 'graphics'.

TIFF: Color or grayscale photographs (halftones): always use a minimum of 300 dpi.

TIFF: Bitmapped line drawings: use a minimum of 1000 dpi.

TIFF: Combinations bitmapped line/half-tone (color or grayscale): a minimum of 500 dpi is required.

If your electronic artwork is created in a Microsoft Office application (Word, PowerPoint, Excel) then please supply 'as is'.

Please do not:

- Supply files that are optimised for screen use (e.g., GIF, BMP, PICT, WPG); the resolution is too low;
- Supply files that are too low in resolution;
- Submit graphics that are disproportionately large for the content.

Color artwork

Please make sure that artwork files are in an acceptable format (TIFF, EPS or MS Office files) and with the correct resolution. If, together with your accepted article, you submit usable color figures then Elsevier will ensure, at no additional charge, that these figures will appear in color on the Web (e.g., ScienceDirect and other sites) regardless of whether or not these illustrations are reproduced in color in the printed version. For color reproduction in print, you will receive information regarding the costs from Elsevier after receipt of your accepted article. Please indicate your preference for color: in print or on the Web only. For further information on the preparation of electronic artwork, please see <http://www.elsevier.com/artworkinstructions>.

Please note: Because of technical complications which can arise by converting color figures to 'gray scale' (for the printed version should you not opt for color in print) please submit in addition usable black and white versions of all the color illustrations.

Figure captions

Ensure that each illustration has a caption. Supply captions separately, not attached to the figure. A caption should comprise a brief title (not on the figure itself) and a description of the illustration. Keep text in the illustrations themselves to a minimum but explain all symbols and abbreviations used.

Tables

Number tables consecutively in accordance with their appearance in the text. Place footnotes to tables below the table body and indicate them with superscript lowercase letters. Avoid vertical rules. Be sparing in the use of tables and ensure that the data presented in tables do not duplicate results described elsewhere in the article.

References

Citation in text

Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full. Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the

publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.

Web references

As a minimum, the full URL should be given and the date when the reference was last accessed. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired, or can be included in the reference list.

References in a special issue

Please ensure that the words 'this issue' are added to any references in the list (and any citations in the text) to other articles in the same Special Issue.

Reference management software

This journal has standard templates available in key reference management packages EndNote (<http://www.endnote.com/support/enstyles.asp>) and Reference Manager (<http://refman.com/support/rmstyles.asp>). Using plug-ins to wordprocessing packages, authors only need to select the appropriate journal template when preparing their article and the list of references and citations to these will be formatted according to the journal style which is described below.

Reference style Text: Citations in the text should follow the referencing style used by the American Psychological Association. You are referred to the Publication Manual of the American Psychological Association, Sixth Edition, ISBN 978-1-4338-0561-5, copies of which may be ordered from <http://books.apa.org/books.cfm?id=4200067> or APA Order Dept., P.O.B. 2710, Hyattsville, MD 20784, USA or APA, 3 Henrietta Street, London, WC3E 8LU, UK.

List: references should be arranged first alphabetically and then further sorted chronologically if necessary. More than one reference from the same author(s) in the same year must be identified by the letters 'a', 'b', 'c', etc., placed after the year of publication.

Examples: Reference to a journal publication:

Van der Geer, J., Hanraads, J. A. J., & Lupton, R. A. (2010). The art of writing a scientific article. *Journal of Scientific Communications*, 163, 51-59.

Reference to a book:

Strunk, W., Jr., & White, E. B. (2000). *The elements of style*. (4th ed.). New York: Longman, (Chapter 4).

Reference to a chapter in an edited book:

Mettam, G. R., & Adams, L. B. (2009). How to prepare an electronic version of your article.
In B. S. Jones, & R. Z. Smith (Eds.), *Introduction to the electronic age* (pp. 281-304).
New York: E-Publishing Inc.

APPENDIX 2: Systematic review protocol
based on York University's Centre for Reviews and Dissemination
Guidance for undertaking reviews in healthcare

Background

- Well established evidence that having a child with a disability impacts on psychological outcomes, e.g. mental health, stress, adjustment, wellbeing.
- Exploration of cognitive factors would be useful clinically, as these are something that could potentially be changed.
- Little research on parents of children with profound and multiple intellectual disabilities (PMID).

Previous similar reviews:

- Narrative review of parental cognitions and r'ship with stress (Hassall *et al.*, 2005)
- Conceptual review of caregiver burden and stress with focus on parents of children with developmental disabilities (Raina *et al.*, 2004)
- Narrative review of lives of people with PMID, including section on parents and carers (Carnaby, 2004).

No systematic reviews conducted in this area.

Review question

What is the relationship between parental locus of control and psychological outcomes for parents who have a child with a disability?

Eligibility criteria

- Published case study, small study, controlled study or non-controlled study
- All types of study design
- Locus of control related to experience of parenting investigated
- Relationship between locus of control and parent psychological outcomes investigated (see below)
- Full-text available
- Article published in English
- All years considered

Population

- Parents or family carers

- At least one child with a physical, developmental or intellectual disability or significant visual or hearing impairment that would constitute a disability

Outcomes

Parent and family psychological outcomes, including:

- Adjustment
- Stress
- Wellbeing
- Mental health (e.g. depression, anxiety)

Planned search strategy

- Keyword searches of online databases (Medline, PsycInfo, PsycArticles, ERIC, CINAHL+, Web of Knowledge), using search terms parent; mother; father; locus of control; disab*
- Manual search of reference lists of papers selected for systematic review
- Manual search of key journals identified as prevalent in reference lists of selected papers

Study selection

1. Abstracts reviewed to see if meet eligibility criteria
2. Full-text of retained studies reviewed to see if meet eligibility criteria
3. Final selection of studies included in methodological appraisal and assessment

Data extraction

The following data was extracted regarding each paper:

- Research/study question
- Study design
- Population(s)/sample(s)
- Measures
- Analyses
- Generalisability of findings

Quality Assessment

- Specific criteria for each dimension
- Scoring categories of well covered; adequately addressed; poorly addressed; not addressed/not reported; not applicable
- Overall assessment of the study to reduce bias (+++; ++; +; and -)

Data synthesis

- Summary of individual study findings and characteristics (data from standardised data extraction form)
- Overall rating and quality ratings for each of the dimensions identified
- Overall summary of state of the literature in this area
- Limitations of available literature
- Areas identified for future research

Dissemination

- Chapter in doctoral portfolio thesis
- Submit for publication

APPENDIX 3: List of search terms used in systematic review

1. parent
2. parental
3. mother
4. mum
5. father
6. dad
7. carer
8. caregiver
9. disability
10. disabilities
11. disabled
12. locus of control

APPENDIX 4: Systematic review data extraction form

General information

Date of data extraction:

Record number (to uniquely identify study):

Author:

Article title:

Citation:

Type of publication (e.g. journal article, conference abstract):

Country of origin:

Source of funding (if known):

Study characteristics

Aim/objectives of the study:

Study design:

Study inclusion criteria:

Study exclusion criteria:

Recruitment procedures used:

Participant characteristics

Age:

Gender:

Ethnicity:

Socio-economic status:

Child disability/syndrome characteristics:

Co-morbidities:

Number of participants in sample:

Intervention and setting

Setting in which the intervention is delivered/research takes place:

Description of the intervention(s) and control(s) (if applicable):

Outcome data/results

Unit(s) of assessment or measure(s) used:

Statistical techniques used:

For each pre-specified outcome:

Measurement tool or method used:

Unit of measurement (if appropriate):

Length of follow-up, number and/or times of follow-up measurements:

For all intervention/experimental group(s) and control group(s):

Number of participants approached/asked to take part:

Number of participants enrolled/took part/returned responses:

Number of participants included in analysis:

Number of withdrawals, exclusions, lost to follow-up:

Summary outcome data e.g.

No. of events or number of participants in each group/condition:

Group/condition mean:

Type of analysis used in study (e.g. group mean comparisons; correlation, etc.):

Results of study analysis:

If subgroup analysis is planned the above information on outcome data or results will need to be extracted for each subgroup:

Details of any additional relevant outcomes reported:

Adverse events:

Other notes:

APPENDIX 5: Quality rating tool for systematic review

	Overview of Quality Criteria Domains	Domain numerical score
1	Research question and objectives	/
2	Sampling	/
3	Design and method	/
4	Statistical analysis	/
5	Quality of reporting	/
6	Generalisability	/
7	Overall assessment of study	/
	Total score	/
	Percentage (based on number of items rated as applicable)	
	Descriptive category (Good $\geq 70\%$, Fair $\geq 50\%$, Weak $< 50\%$)	

1. Research question and objectives

1.1 The study addresses an appropriate and clearly focused question, drawn from a theoretical model or previous research.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

2. Sampling

2.1 The characteristics of the participants are representative of the group being studied.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

2.2 The study indicates how many of the people asked to take part did so, in each of the groups studied. Also, if applicable, how many dropped out.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

2.3 The exact nature and severity of the child's disability/impairment and its impact on every day family and child functioning is described.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

3. Design and method

3.1 The constructs/variables under investigation are clearly defined and operationalised.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

3.2 Variable measurement method is appropriate and demonstrates validity and reliability.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

3.3 Confounding variables that may have influenced the results are taken into account.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

4. Statistical analysis

4.1 Statistical analyses are fully reported and appropriate.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

5. Quality of reporting

5.1 Reporting of method, analyses and results are sufficiently detailed to allow their replication or justification.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

6. Generalisability

6.1 The findings could be generalised to similar populations.

Well covered = 3	
Adequately addressed = 2	
Poorly addressed = 1	
Not addressed/not reported = 0	
Not applicable = 0	
Other comments	

7. Overall assessment of study

7.1 A judgement of the overall quality of the study.

++ Good to excellent = 3	
+ Adequate to good = 2	
- Poor to adequate = 1	

APPENDIX 6: Participant Information

About the survey

Dear Parent / Carer

I work with children and young people with profound and multiple disabilities and their families. Some people use other labels to describe profound and multiple disabilities.

I am referring to children and young people under 19 who have more than one disability, including:

- a profound learning disability
- significant communication difficulties
- sensory impairment (e.g. hearing, vision)
- physical disabilities and mobility problems
- complex health needs (e.g. enteral/parenteral feeding, ventilation/CPAP, etc)
- mental health difficulties

These individuals need high levels of support with most aspects of daily life. This criteria comes from Mencap's leaflet on the definition of profound and multiple disabilities (accessed at <http://www.pmlnetwork.org>).

Families respond in different ways to having children with profound and multiple disabilities. For some this can be a positive and enriching experience, for others it can bring a number of challenges. I have met some families of children and young people with profound and multiple disabilities who are struggling with the emotional aspects of caring for their child, as well as the more practical demands. I have also met some families who have had positive experiences.

I would like to understand more about the process of adapting to having a child with profound and multiple disabilities and how psychologists and other professionals may be able to support this. I would be very grateful if you could complete this online survey as part of my research. The research project has had ethical approval from the School of Health in Social Science at the University of Edinburgh and is being supervised by Dr Helen Downie and Dr Karen McKenzie.

If your child has more than one parent (or carer with parental responsibility), it would be really useful if you could each fill in the online survey. Please direct other parents/carers of children with profound and multiple disabilities to it at:

<http://www.survey.ed.ac.uk/parentcarerwellbeing>

The online survey contains different questions, which should take around 30 minutes to complete in total. There are no right or wrong answers - I am interested in what you think

and how you feel. All your answers will be anonymous and you will not be identifiable. If you have concerns that information we are asking for will identify your child or your family, you do not have to provide this information.

Some of the questions might make you feel or think about things that are difficult and/or upsetting. We recognise that having a child with profound and multiple disabilities can have both negative and positive aspects. If you want to talk about this or would like some advice about seeking formal support for this, please contact me using the details on the next page. If you are happy to take part and complete the questionnaire, the next page has details about what this will involve, followed by the questions.

Thank you for your time.

Fleur-Michelle Coiffait
Trainee Clinical Psychologist

Clinical & Health Psychology
School of Health in Social Science
University of Edinburgh
Old Medical School
Teviot Place
Edinburgh EH8 9AG
Tel: 0131 651 3972
Email: parentwellbeingresearch@gmail.com

What will participation involve?

Taking part in this research will involve answering the questions contained in this booklet and returning your completed questionnaire using the postage paid envelope provided. If you would prefer to complete the questionnaire online, please visit:

<https://www.survey.ed.ac.uk/parentcarerwellbeing>

What will happen to this information?

The information will be anonymous and will be used only for the purpose of this research. The overall findings will be written up and shared with other researchers and professionals to help ensure that families' needs are better understood. We are not asking for information that could be used to identify you or your family. If you have concerns that information we are asking for will identify your child or your family, you do not have to provide this information.

What if I do not want to take part?

You do not have to complete the questionnaires, only complete them if you want to. Your decision to participate or not in this research *will not* affect any services you may be receiving.

What if I change my mind?

If you change your mind once you have returned your questionnaire, you have the right to withdraw your information from the study. Please make a note of your unique participant ID (on the top right corner of the cover page) so that we are able to identify and destroy your data securely, if you decide to withdraw your information after submitting your responses.

What if I want to talk to someone?

If you have any questions about the study, you can contact me by

Email: parentwellbeingresearch@gmail.com

Telephone: 0131 651 3972

I can also help you find out about further support if you feel this would be necessary.

By completing and returning this questionnaire, I confirm that I am aware of my right to withdraw my information and I give my informed consent for my responses to be used for the purpose of this research study.

APPENDIX 7: Organisations that supported the study

The author would like to extend a huge thank you and her gratitude to the following organisations for their support in publicising the study:

Capability Scotland Stanmore House School, Lanark
Cerebra
Challenging Behaviour Foundation
Enable Scotland
Epilepsy Scotland
Families Magazine
Foundation for People with Learning Disabilities
Genetic Alliance UK
Kindred Scotland
LD Health Network
Parents Own
PAMIS
Scottish National Managed Clinical Network for Children with Exceptional Healthcare Needs
PMLD Link
Rare Disease UK
Royal Blind School, Canaan Lane Campus, Edinburgh
Shine UK
Special Needs Parents Association
Syndromes Without a Name (SWAN) UK
Together for Short Lives
Unique
Voice of Carers Across Lothian (VOCAL)

Additional email lists, forumd and social media sites used to advertise the study

BPS Division of Clinical Psychology Paediatric Psychology Network email list
ClinPsy.org.uk online forum
Intellectual Disability Research UK JISCmail list
LD Health Network forum
Psychology Postgraduate Affairs Group (PsyPAG) JISCmail list
Twitter (<http://www.twitter.com/PMLDresearch>)

APPENDIX 8: Parental Locus Of Control Scale - revised version (PLOC-R)

(Lloyd & Hastings, 2009; *cf.* Campis, Lyman, & Prentice-Dunn, 1986 for original version)

Many parents who have a child with special needs believe that particular child has had a special effect on them and on other members of their family. What effect do you believe your child with a disability has had on you and other members of your family?

Read each statement and indicate the one response that best describes how much you agree or disagree with each statement using the following answers:

Note: Items followed by (R) are reverse scored

Parental Efficacy Subscale	Strongly Disagree	Disagree	Not Sure	Agree	Strongly Agree
What I do has little effect on my child's behaviour.	1	2	3	4	5
When something goes wrong between me and my child, there is little I can do to correct it.	1	2	3	4	5
If your child throws tantrums no matter what you try, you might as well give up.	1	2	3	4	5
My child usually ends up getting his/her way, so why try?	1	2	3	4	5
No matter how hard a parent tries, some children will never learn to be responsible.	1	2	3	4	5
It is not always wise to expect too much from my child because many things turn out to be a matter of good or bad luck anyway.	1	2	3	4	5
When my child gets angry I can usually deal with him/her if I stay calm. (R)	5	4	3	2	1
When I set expectations for my child, I am almost certain that I can help him/her meet them. (R)	5	4	3	2	1
Parental Responsibility Subscale	Strongly Disagree	Disagree	Not Sure	Agree	Strongly Agree
When my child is well-behaved, it is because he/she is responding to my efforts.	1	2	3	4	5
Parents who can't get their children to listen to them don't understand how to get along with their children.	1	2	3	4	5

My child's behaviour problems are no one's fault but my own. (R)	5	4	3	2	1
Capable people who fail to become good parents have not followed through on their opportunities. (R)	5	4	3	2	1
Children's behaviour problems are often due to mistakes their parents make.	1	2	3	4	5
Parents whose children make them feel helpless just aren't using the best parenting techniques. (R)	5	4	3	2	1
Most children's behaviour problems would not have developed if their parents had had better parenting skills.	1	2	3	4	5
I am responsible for my child's behaviour. (R)	5	4	3	2	1
The misfortunes and successes I have had as a parent are a direct result of my own behaviour.	1	2	3	4	5
<i>Child Control of Parent's Life Subscale</i>	Strongly Disagree	Disagree	Not Sure	Agree	Strongly Agree
My life is chiefly controlled by my child.	1	2	3	4	5
My child does not control my life. (R)	5	4	3	2	1
My child influences the number of friends I have.	1	2	3	4	5
It is easy for me to avoid and function independently of my child's attempts to have control over me. (R)	5	4	3	2	1
I feel like what happens in my life is mostly determined by my child	1	2	3	4	5
<i>Parental Belief in Fate/Chance Subscale</i>	Strongly Disagree	Disagree	Not Sure	Agree	Strongly Agree
Being a good parent often depends on being lucky enough to have a good child.	1	2	3	4	5
I'm just one of those lucky parents who happened to have a good child.	1	2	3	4	5
I have often found that when it comes to my children, what is going to happen will happen.	1	2	3	4	5

Fate was kind to me—if I had had a bad child I don't know what I would have done.	1	2	3	4	5
Success in dealing with children seems to be more a matter of the child's moods and feelings at the time rather than one's own actions.	1	2	3	4	5
Neither my child nor myself is responsible for his/her behaviour.	1	2	3	4	5
In order to have my plans work, I make sure they fit in with the desires of my child.	1	2	3	4	5
Most parents don't realise the extent to which how their children turn out is influenced by accidental happenings.	1	2	3	4	5
Heredity plays the major role in determining a child's personality.	1	2	3	4	5
<i>Parental Control of Child's Behaviour Subscale</i>	Strongly Disagree	Disagree	Not Sure	Agree	Strongly Agree
I always feel in control when it comes to my child. (R)	5	4	3	2	1
My child's behaviour is something more than I can handle.	1	2	3	4	5
Sometimes I feel that my child's behaviour is hopeless.	1	2	3	4	5
It is often easier to let my child have his/her way than to put up with a tantrum.	1	2	3	4	5
I find that sometimes my child can get me to do things I really did not want to do.	1	2	3	4	5
My child often behaves in a manner very different from the way I would want him/her to behave.	1	2	3	4	5
Sometimes when I'm tired I let my children do things I normally wouldn't.	1	2	3	4	5
Sometimes I feel that I do not have enough control over the direction my life is taking.	1	2	3	4	5
I allow my child to get away with things.	1	2	3	4	5
It is not too difficult to change my child's mind about something. (R)	5	4	3	2	1

APPENDIX 9: Positive Gain Scale (PGS)

(Pit-ten Cate, 2003)

The following statements focus on your own and your family's experiences of having a child with intellectual disability (ID).

Please respond to all questions by indicating the answer that best describes how you feel.

	Strongly Agree	Agree	Not Sure	Disagree	Strongly Disagree
Since having this child I feel I have grown as a person.	1	2	3	4	5
Having this child has helped me to learn new things / skills.	1	2	3	4	5
Raising this child helps putting life into perspective.	1	2	3	4	5
Since having this child, my family has become closer to one another.	1	2	3	4	5
Since having this child, my family has become more tolerant and accepting.	1	2	3	4	5
Since having this child I have become more determined to face up to challenges.	1	2	3	4	5
Since having this child I have a greater understanding of other people.	1	2	3	4	5

APPENDIX 10: Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS)

(Stewart-Brown *et al.*, 2009; Tennant *et al.*, 2007)

Below are some statements about feelings and thoughts. Please tick the box that best describes your experience of each over the last 2 weeks.

	None of the time	Rarely	Some of the time	Often	All of the time
I've been feeling optimistic about the future	1	2	3	4	5
I've been feeling useful	1	2	3	4	5
I've been feeling relaxed	1	2	3	4	5
I've been feeling interested in other people	1	2	3	4	5
I've been dealing with problems well	1	2	3	4	5
I've been thinking clearly	1	2	3	4	5
I've been feeling good about myself	1	2	3	4	5
I've been feeling close to other people	1	2	3	4	5
I've been feeling confident	1	2	3	4	5
I've been able to make up my own mind about things	1	2	3	4	5
I've been feeling loved	1	2	3	4	5
I've been interested in new things	1	2	3	4	5
I've been feeling cheerful	1	2	3	4	5

Warwick-Edinburgh Mental Well-being Scale (WEMWBS)

© NHS Health Scotland, University of Warwick and University of Edinburgh, 2006

All rights reserved